A STUDY OF THE PSYCHOLOGICAL CONCOMITANTS OF A CHRONIC ILLNESS IN CHILDHOOD.

ANN M. CULL

Ph.D.
UNIVERSITY OF EDINBURGH
1974

CONTENTS.

VOLUME II

LIST OF APPENDICES

ACKNOWLEDGMENTS

Several people have given their help and advice during the course of this study and to all of them I should like to extend my warmest thanks.

In particular, I should like to thank my two supervisors for their guidance. Mr. D. McMahon has been a most kind and wise mentor throughout my postgraduate career; Dr. W.M. McCrae originally saw the need for research in this area and, having appointed me to carry it out, he gave most freely of his time and experience at all stages of its development. I am deeply grateful to them both for their teaching, their help and their encouragement.

I am indebted to the Cystic Fibrosis Research Trust by whose financial support this study was made possible.

I should also like to thank Prof. D.H. Hutchison, Dr. R.H. Shanks and Dr. M. Fraser who were kind enough to allow me to include some of their patients in the study. I am grateful too, to the Records Department of the Royal Hospitals for Sick Children in Edinburgh and Glasgow for their help in tracing patients.

Mr. Neil Hamilton-Smith provided invaluable assistance in the computer analysis of the data and I should like to express my appreciation of the endless patience with which he met my ever more urgent requests for computer time.

The preparation of the thesis has been something of a family affair and my warmest thanks go to my father for providing typing facilities and to my mother for her painstaking proof-reading.

Mrs. E. Stevenson typed the manuscript and I am most grateful to her for her hours of work. My husband endured with great patience the trials of having a 'liberated' wife while this thesis was in preparation and for his efforts at housewifery as well as for his continuing support and encouragement, my warmest thanks are due.

Finally, and most important of all, my deepest gratitude goes to those families who allowed me to come into their homes and who gave so much thought and time to their contributions to this study. As a token of my appreciation of their help this thesis is dedicated to them and their chronically ill children.

A B S T R A C T

Cystic fibrosis is an inherited and potentially fatal disease which affects, primarily, the functioning of the lungs and of the pancreas. Although there is no cure for cystic fibrosis, modern medicine has achieved a measure of control over its progress and the prognosis for affected children has thereby improved to the point where those who would previously have died in their first year are now surviving into adolescence and young adulthood. Thus we may now begin to regard cystic fibrosis as a chronic disease of childhood.

British children with this disease have not previously been studied from a psychological point of view but, in general, studies of the concomitants of other chronic physical illnesses or disabilities have described the afflicted children as being particularly vulnerable to social and psychological difficulties. Although cystic fibrosis is rather different from many of these disorders previously studied, being, in most cases, neither disfiguring nor disabling, investigations of C.F. children conducted in the United States indicated, nevertheless, a disturbingly high incidence of psychopathology among these children and their parents. These studies had some important shortcomings and a need for the clarification of the nature of the social and psychological concomitants of cystic fibrosis was apparent. This study sought to meet this need by describing the influence of the disease on the lives of a sample of British children and their families.

Fifty families with 58 C.F. children assisted in this

enquiry. Data was gathered by interviews and by psychometric tests, carried out on successive occasions in the families' own homes in Central Scotland.

Since parents play a central role in the treatment of their C.F. children their attitudes to the medical aspects of the problem were of primary importance and detailed information was collected to describe their views of the management of their child's diagnosis and his long-term care. The financial and social consequences for the family as a whole were considered and the burden placed on parents was examined in terms of the parental, marital and personal aspects of their lives. The influence of the disease on the intellectual, social and emotional development of the affected children was explored and the children themselves contributed to this stage of the investigations. Teachers' assessments of the children's behaviour at school added a further dimension to the study. Finally the implications of the situation for the other health children in the family were briefly reviewed.

The findings were evaluated with reference to a control study of the concomitants of coeliac disease. This condition is similar to cystic fibrosis in that it is an astigmatic and incurable chronic disease which also demands a high degree of family involvement in the child's treatment, but it does not have a life-threatening prognosis. A sample of coeliac children was selected to correspond to the C.F. sample in several important characteristics and they and their families took part in an investigation which followed almost exactly the design of the original study.

The results of these two studies are compared and some problems of management, particularly of cystic fibrosis are thus highlighted. Some possible means of alleviating these difficulties are discussed. Attention is drawn to the potentially disruptive influence which the chronic disease of a child may have on all the members of his family, especially if his disease has a grim prognosis. Family communications and parents' attitudes to their sick child are identified among a number of key areas in which such disruption can have far-reaching consequences for the child's personal development. The study attempts to show how this tendency for existing physical damage to become compounded with social and emotional problems might be combatted.

The findings are encouraging in that the gross psychopathology of the American studies was not evident in this sample. The study concludes with the hope that renewed awareness of the social and psychological concomitants of cystic fibrosis might enable those who care for these children and their families to improve the personal outlook for them by as much as the medical profession has improved the clinical prognosis.

SECTION I

BACKGROUND TO THE STUDY

Before embarking on a study of the social and psychological aspects of any specific chronic disease process, some consideration should be given to the broader background of knowledge and ideas already prominent in the more general aspects of this field, i.e.

A. to the major theoretical principles and positions popularly held in the psychology of chronic illness and disability.

B. to the methodological problems that need to be overcome in the verification of these theories, and

C. to the literature reporting studies of disability in general and of other specific ailments, which may suggest methodology, hypotheses or findings relevant to this problem.

Only then is the way prepared for consideration of the issues which relate specifically to the disease in question. Thus the background specifically required for such a study of cystic fibrosis is discussed in the two remaining sub-sections, D and E:

D. in which the aetiology, symptoms and treatment of cystic fibrosis are explained in lay terms, and

E. in which the literature reporting studies of cystic fibrosis children and their families is discussed.

A. The Theoretical Basis

In the history of psychology, several attempts have been made to correlate the anatomical characteristics of man with his behaviour, ranging from the humoral notions of Hippocrates, the "Father of Medicine", to the phrenology of Gall. In more recent times elaborate systems have been devised to relate personality types to physique.

These approaches offer little relevant information to clarify the problem in hand so that only the four major contemporary theoretical standpoints are discussed here.

1. Adler's Individual Psychology

Adler departed from classical psychoanalytic theory in his emphasis on individual uniqueness and on social, rather than sexual,urges. The most notable of the concepts which he introduced, which are relevant here, are those of striving for superiority, inferiority feelings, compensation and style of life.

For Adler, all behaviour was goal-directed, towards goals that were primarily social in character, and it was driven, by an innate striving toward superiority or self actualisation. To understand how the particular forms of striving for superiority come into being in the individual, one must consider too, Adler's concept of inferiority feelings.

Early in his career Adler introduced the notions of organ inferiority and overcompensation in an attempt to account for afflictions to particular body regions. Recognising that there was often in these cases a predisposing inferiority in one region

of the body owing to a developmental or genetic abnormality, Adler observed that the person with the defective organ often tries to compensate for the weakness by strengthening it through intensive training.

Later Adler developed the more general thesis that feelings of inferiority arose from a sense of imperfection in any sphere of life. The individual was then conceived of as developing a definite mode of attempting to achieve perfection in some realm and this was called his style of life.

The origins of both the striving for superiority and the style of life were firmly rooted in early childhood. The child, Adler said, has a feeling of inferiority relative to his parents, and to the world in general.

...."Thus the child arrives at the positing of a goal, an imagined goal of superiority, whereby his poverty is transformed into wealth, his subordination into domination, his suffering into happiness and pleasure, his ignorance into omniscience and his incapacity into artistic creation." Alfred Adler, 1927.

Thus at a very early stage a style of life begins to develop which is the individual's own unique way of striving toward the common goal of superiority. Throughout the rest of his life he selects and assimilates those experiences which fit his style of life, ignoring those which do not. New ways of expression are attained and the old discarded, but the style of life remains the same.

Now Adler was particularly interested in the kinds of early influences predisposing a child to a faulty style of life and he identified physical and mental weaknesses among the principal causes. The longer, and more intensely, insecurity and dependency were

felt, the greater the suffering imposed, and the more acutely the person became aware of life's misfortunes so then, he said, the higher would the goal of superiority be placed and the more rigidly would it be pursued. Thus Adler saw children with mental or physical infirmities as likely to feel inadequate in meeting the tasks of life and to consider themselves failures. He did point out, though, that understanding, encouraging parents could help the children to compensate for their inferiorities and to transform weakness into strength.

Striving for superiority and seeking to compensate for inferiority are natural, indeed, essential processes in Adler's schema, but they can be distorted and, in his analysis, Adler interpreted the exemptions and privileges of illness and disability as providing a substitute for the original goal of superiority, in the opportunities they afford for the manipulation and domination of others. Closely akin to this, Adler spoke out against the pampering of children which he also felt nurtured despotic tendencies.

Although the Adlerian approach is, in some respects, an appealing one, there are several difficulties particularly in the application of the concepts of inferiority and compensation.

The relation between disability and feelings of inferiority is not as inviolate as Adler assumed. In a fairly comprehensive review of the literature Wright (1960) could find little convincing evidence of inferiority feelings among the disabled, although this was indeed a characteristic frequently assigned to them by others. There was, however, some evidence that disability could contribute to a person's striving for superiority in the

terms described above, by allowing him to behave in ways that would not otherwise be acceptable.

There is a shortage of rigorous studies, and a plethora of anecdotes and folk lore in relation to compensation and this has created a problem of definition. Wright (1960) sees compensation as leading to a shift in values. This is a simple conceptualisation which is meaningful, not only in the study of disability, but it does not accommodate the shift of attention which compensates for sensory loss, by selecting previously unused cues available on the remaining open channels. Although an interesting problem, the matter of compensation is not of primary importance to the problem in hand and will not be pursued in detail here.

Thus, for the lack of substantive evidence to back it, we have to conclude that Adler's theory offers us little means of understanding the general relationship of disability to behaviour.

2. Schilder's Theory of Body Image

The concept of a body image which may be disrupted by chronic illness or disability has become popular, at least among research workers. The 'postural schema' of the body as described by Sir Henry Head, laid the foundations for this concept which has since been elaborated, within a more psychoanalytic framework, by Schilder.

All sources of sensation and spatial relationships of the body contribute to this "most fundamental of all human experience", the body image. The perception and representation of the body image is not necessarily conscious, although much of behaviour and perception are said to be determined by it. Beyond

this it seems a very vague construct which Schilder himself has clearly found difficulty in expressing.

Of the effects of disease in general on the body image, Schilder said, "There arises the problem of organic disease in connection with the postural model of the body. Organic disease provokes abnormal sensations; it immediately changes the image of the body, partly the picture side of it and partly the libidinous investment. These sensations immediately become a part of the general attitude and experience of the individual." Paul Schilder, 1950.

This has had the effect of stimulating a tremendous research effort into establishing that a whole gamut of chronic illnesses do indeed lead to some change in the patient's inferred body image. Unfortunately little use seems to have been made of this finding. It was suggested that, if a discrepancy arose between the person's actual physique and this body image, this could lead to emotional conflict, fantasy and delusional processes, but no study was found which tested the relationship between body and personal adjustment.

The most promising application of the body image concept seems to lie in work with amputees and in explaining phantom sensations, or, as suggested by McDaniel, (1969) it may prove a useful addition to the more global self-concept. As it is presently defined, the hypothetical construct of body image seems limited in its utility, with scant contribution to make to our study of cystic fibrosis.

3. Parson's Social Role Theory

Talcott Parsons made an important contribution to our understanding of psychological responses to illness and disability in his essay on "Definitions of Health and Illness in the Light of American Values and Social Structure." (1958)

He began with a number of basic assumptions of role theory. First describing an individual's role in terms of his performance of various, differentiated tasks within his own social system, Parsons went on to identify health as the optimal state for the performance, to capacity, of these valued tasks. Conversely then, illness or disability ~~are~~ is seen to produce incapacity and thus to limit or inhibit the performance of accustomed tasks. Reverberations may then be felt in the family, as the most fundamental social system to which an individual may belong, where role patterns may be disrupted or, depending on the permanence of the disability, completely reorganised. Rehabilitation is then the term that is used to describe any treatment or service which is designed to restore or, at least, to optimise the person's capacity for appropriate role performance.

As the title of Parsons' essay suggests, these assumptions take on their full meaning only when they are viewed in cultural perspective and he describes the American value system, in relation to physical health, most lucidly.

It becomes clear that health is the natural state and that to be ill is to deviate from the norm. Although the sick or disabled are not, then, held to be accountable for their deviant position, they are not entirely exempt from social role requirements, and the sick role itself is a demanding one, as Parsons relates.

In American society as described by Parsons, health and

education are valued, as the basis for independent achievement and economic productivity. The capacity for achievement is said to be developed through education and protected or restored by the health services.

The social role approach to the study of disability would seem to be a fruitful one, although there has not been a great deal of research to augment Parsons' seminal work. Perhaps this is because his system is not so amenable to the study of individual psychological effects in illness and disability which have been the interest of so many researchers. Nevertheless his account of the sick role and its impact on family functioning have highlighted new aspects of our problem in relation to cystic fibrosis.

4. Barker and Wright's Somatopsychology

Barker (1960) was very critical of approaches to the relationship between psyche and soma which invoked "unsystematic recondite processes" e.g. organ inferiority, body image or which concentrated on the technological problems for practitioners, e.g. in counselling, in education, and he welcomed the new importance given to the phenomenal and instrumental significance of physique by Wright's book on "Physical Disability - A Psychological Approach."

In fact she was continuing along the lines which he himself had laid down in 1953 when he defined somatopsychology as the study of - "those variations in physique that effect the psychological situation of a person by influencing the effectiveness of his body as a tool for actions or by serving as a stimulus to himself or others." Broadly then, somatopsychologists see physique and behaviour as being closely

interrelated and interdependent.

In relation to physical disability, Wright (1960) was careful to point out that the social aspects of the condition could be as handicapping as the physical. Thus the combination of behavioural incapacities and social rejection were held to place the disabled in a subordinate position, analogous to that of minority groups, where many goals became inaccessible. This idea introduced the concept of devaluation which embraced not only the Adlerian notion of inferiority or feelings of lowered self esteem, but also the attitude of others to the disabled person. These latter attitudes, usually of overprotection or rejection, have since been the focus of a flood of research interest and some relevant studies will be discussed later. These attitudes are based on the prejudged helplessness and dependency of the disabled person who then may reflect this devaluation of his own position. The whole situation may become still more distorted by the phenomenon of "spread", i.e. the disability is not confined to the actual limits of the impairment, so that the individual may be viewed, and indeed may come to view himself, as being incapacitated in additional ways.

Another interesting principle of behaviour which was suggested by the somatopsychologists and quickly taken up by several others, was that of "mourning" or depression, and the acceptance of loss. The aspect of this reaction which is of concern in this study relates, not to the patient, but to his family in their acceptance of his affliction. Nevertheless it is of interest to consider the theory put forward here by

Barker et al and by Wright. They suppose mourning for a lost function or part to be inevitable and indeed desirable, for they equate it with the realisation of the loss, the sine qua non of successful acceptance of the disability.

Wright (1955) described the mourning reaction as being characterised by despair, depression and emotional withdrawal then, following the reconstruction of the self concept, altered to accommodate the physical change, the mourning abates and a sense of personal worth is re-established.

Although elements of this somatopsychological approach have become popular among social workers and those concerned with the rehabilitation of the disabled there has been a marked lack of research evidence to substantiate them.

As early as 1953, Barker pointed out a number of methodological pitfalls awaiting research in this field and we shall take time to consider these in relation to this study in a later chapter. Seven years later, in an assessment of the development of somatopsychology up to 1960, Wright observed that "inconsistency is fairly typical of the status of many of the findings." This would seem to hold true even today.

Thus somatopsychology is more accurately described as a guide to further study than as a body of knowledge. The suggested principles of the psychological aspects of physical disability are worthy of further consideration as are the many intervening variables which Wright has listed; nevertheless the whole depends heavily for its illustration on a collection of anecdotes and as Wright herself has said...."There is much room for further conceptual clarification." (Wright 1960)

A review of the major contemporary theories involving the psychological aspects of chronic illness and physical disability is, then, frankly disappointing. Among a welter of concepts there is a striking paucity of substantiating fact. In addition there is still an abundance of unfounded opinion and folklore which makes the task of sorting out the facts the more formidable. For the present the most promising approach would seem to be an eclectic one, incorporating only those elements which have been validated by adequate research.

Before reviewing the literature for relevant information we shall examine some of the methodological difficulties inherent in this work in order to establish criteria by which to assess the reported research.

B. Methodological Considerations

Barker (1953) discussed the methodological shortcomings of attempts to validate the somatopsychological viewpoint but his critique can be usefully applied to many of the studies in this field and should certainly be borne in mind in this study of cystic fibrosis.

1. Isolating relationships

Barker wrote of the difficulty of disentangling and distinguishing between the several links between physique and behaviour, e.g. genetic, endocrine, neural and psychosomatic as well as somatopsychological links, even when the effects of other relationships were allowed for by the use of adequate controls, and by the judicious selection of appropriate research techniques, Barker pointed out that only relative purity of the physical-behavioural relationship could ever be attained. This limitation should be acknowledged in the terms of the research being reported.

2. Selection of representative subjects

Persons with deviations in physique are not necessarily randomly distributed in the general population. It behoves the researcher first to determine the distribution of his subjects in the general population in order to obtain a representative sample. However obvious this criterion, it is not easily satisfied. The onus is then upon the researcher to provide adequate controls.

3. Controls

The adequacy of the control group chosen has to be judged from each investigation on its own merits, bearing in mind the physical deviation involved, and the kind of behaviour being studied. That there should be some control study is beyond dispute since, in this field, there is a large number of intervening variables which cannot otherwise be eliminated. Even so, it is rarely possible to secure adequate equivalence of irrelevant variables.

Although Barker quotes twin controls as being among the best of the available possibilities in this field, the author feels that the exclusive selection of twins violates too drastically the requirement of the representativeness of subjects. Self-Controls, as used in a before-and-after paradigm, might be of interest in relation to acquired disabilities, although advance information cannot often be available of impending illness or disability, and certainly this method is not applicable to the study of chronic illness.

The options are thus restricted to a selection of controls from more or less disabled groups or indeed from normal subjects.

Except in a few isolated cases it has proved difficult to demonstrate comparability when non-disabled subjects are used so that the range of acceptable subjects for control studies in the psychology of disability is indeed limited.

4. Assessing physical variation

A problem exists in evaluating the research reported in

the literature, in the lack of standardisation of description and measurement of different kinds of physical illness and disability. Without knowing the nature and degree of the disability it is very difficult to assess the comparability of the findings. It is of interest in this context however that the somatopsychological viewpoint would suggest that the type and degree of the disability should be of little significance.

5. Data on the subject's situation

It is evident from the research that has accumulated that it is necessary to point out that a more global account of the person's life situation needs to be given in concrete detail for the research worker's findings about him to be interpreted correctly.

6. Describing behaviour and personality

Difficulties in studies of personality and everyday behaviour are well known in the main stream of psychology where research on normal subjects has been beleaguered by doubts about the reliability, validity and appropriateness of the tests to be used. These issues come further into question when the tests are applied to disabled subjects.

Barker (1953) found a total absence of research in the literature which was adequate on all six counts. He resigned himself to making the best of the inadequate data and imperfect methodology available at that time. Only a careful review of the literature to date will tell whether we, too, must be so resigned.

C. Review of the Literature. Part 1. Concomitants of physical disability and chronic illness.

Until fairly recently research workers in the psychology of disability and chronic illness have had two major preoccupations - the personal adjustment of the affected individual and the attitudes of others to him and his disability. Initially, these were studied separately.

The matter of how individuals respond and adjust to the occurrence of permanent physical damage due to disease or injury is, to some extent, the most important aspect of rehabilitation in its broadest sense. The success of any remedial measures in terms of long term benefit to the individual is said to depend on his personal adjustment. It is important then to discover the progress which research has made in discovering the status of the emotional adjustment of the physically disabled.

Another of the more important subjects for study in the relationship between psychological variables and physical disability is that of attitudes. Studies here show the development of two approaches to the attitudes of the non-disabled to the disabled. These are the 'prejudice' or 'minority group' and the "body-concept' views. The research evidence to support these views will be considered briefly.

As the importance of family relationships and particularly of parental attitudes has grown, so these two research interests have amalgamated in a plethora of studies pointing out the importance of parent-child relationships for the adjustment of the disabled child. These studies are important for this study, and will be reviewed in detail.

Practical problems seem to have been probed in isolation by the professionals specifically concerned with social, financial, educational and vocational issues for there have been complaints that these researchers have been blinkered to any problems other than those mundane problems of day to day management. (McDaniel,1969) It is unclear what has become of the findings of some of these early studies. There are few reports in the literature which consider psychological aspects of disease against the backdrop of the practical problems it poses, yet the reports must have escaped even the professional literature for there is still concern expressed for the need to get the available research into the educational process and out to the practitioner. In any event there is a marked lack of appreciation of practicalities in the early part of this review.

In 1963 a problem of this nature, but in another context, was apparent to Runciman when he wrote: "A significant advance has still to be made in the social sciences in the large, uncertain and difficult areas where psychological factors interact or overlap with social....The problem which therefore confronts the social scientist is that of trying to formulate, in a particular area of behaviour, some general propositions about the conditions under which a proportional influence should be assigned to each type of variable."

Essentially this is a reformulation of the methodological difficulties already acknowledged of isolating relationships and considering the subject's situation. The call for the integration of different disciplines to bring a more comprehensive approach to problems in human behaviour has stimulated further research in the study of disability. More recent reports show that the total

situation, not only of the afflicted individual, but also of his family and their social environment, has begun to be deemed worthy of systematic study and of a place in the scientific literature. Such studies have much information to offer and will be reviewed in some detail.

It should be noted that a large majority of studies reported both in this review, and in the literature as a whole, refer to physical disabilities which are to some extent handicapping, disfiguring, and stigmatising. The significance of the differences between such conditions and our topic, cystic fibrosis, will also be discussed.

For the moment let us consider the studies of the psychological aspects of chronic illness and disability which have been reported in the literature. These are best discussed under 4 major headings;

(a) Early studies of individual adjustment to physical disability

(b) Studies of attitudes and disability

(c) Parent-child relationships

(d) Modern studies : The global approach to chronic illness

(a) Early studies of individual adjustment to physical disability (1928 - 1960)

It is interesting, amid so voluminous a literature, that one of the earliest studies should have retained its significance when many subsequent works have been discredited. This is certainly the case in the study by Allan and Pearson (1928) of the emotional problems of physically handicapped children.

Following on from Adler's theory, the prevailing view at that time was that organic or physical defect, by its interference in the child's intellectual, social and emotional development, would inevitably lead to a crippled or disordered personality.

Allan and Pearson believed that physical disabilities in the early years of life affected personality development largely in accordance with the effect which they had on the relationship between the parents and the child. They identified three kinds of parental attitude to crippled children which they considered harmful, namely:

1. Inconsistent behaviour involving careful provision for physical care but resentment of the burden thus imposed.
2. Outright rejection.
3. Overprotection.

'Crippling of the personality' was seen as a more serious menace to the future happiness of the individual than a very marked physical handicap so that consideration of parent-child relations was deemed essential. Desirable parent behaviour was accorded when parents showed an objective and realistic acceptance of the child's limitations, with sufficient care and affection to provide security for the child, but not so much as to lead to emotional dependency.

Although these conclusions were based on a highly subjective study of the life histories of children suffering from a variety of handicaps, they will be reiterated in much of the so called objective research that is forthcoming.

Meng (1938) agreed that it is the treatment which these children receive which makes maladjustment probable and he recorded from his interview and questionnaire data that false sympathy, indulgence, severity and even neglect were unfortunately common among parental attitudes. Meng takes a psychoanalytic view in his suggestion that there are three ways for parents and teachers to go about their chief task of diminishing the child's anxiety. These are:-

1. to bring to consciousness the child's blame of his parents, thereby avoiding the problems that arise when these feelings are repressed:
2. to encourage play in order that the child should not, because of his disability, develop prematurely and superficially into a little adult:
3. to help the child to set himself realistic goals.

Kammerer (1940) studies the personality traits of fifty children suffering from scoliosis and thirty children suffering from osteomyelitis. In what proved to be one of the most adequate studies reported in the early literature, he concluded that where personality problems had been found they reflected the number and severity of problems confronting the child, not simply the immediate repercussions of his disability. He did, however, add support to Adlerian notions of the evils of pampering, for he recorded 'spoiling' in 64% of the cases and described these children as being less well adjusted than the 36% who were not spoiled.

Gates (1946) gathered autobiographical data and an exhaustive battery of test results from a sample of young adults

with physical disabilities and from a normal control group. He endorsed these views about the importance of personal and social relationships, particularly within the home environment, for the adjustment of the child. He also seems to have been one of the first to appreciate that the presence of the disabled child in the home could affect the adjustment of all the family members.

The emphasis in the question of adjustment to disability then began to shift. Cruikshank (1951) tried to evaluate the role of fear in the children's adjustment, comparing the responses of children suffering from a variety of conditions - mainly poliomyelitis, heart disease and cerebral palsy - with the responses of normal children. On a sentence completion test, he found the handicapped children to have more fears and feelings of guilt than the children of normal physique and he suggested this might be important for their adjustment.

This paved the way for a greater appreciation of the importance of the individual's interpretation of his own disease. In 1952 the United States Children's Bureau published a report which was intended to outline for professional workers the emotional needs of children suffering from all types of disability. It pointed out the hitherto neglected fact 'that whatever the reality of the disease the child is inclined to interpret it as punishment for his deeds and thoughts or to believe it to be a result of parental rejection or to think of it as a sign that he is an outcast. Individual interpretation will be shaped by the unconscious meanings that disease and handicap have for the child.'

Barker and Wright (1952) developed these notions further. Conceding that the adjustment problems of the disabled are in many respects unique to each individual, they moved that there were,

nevertheless, certain common psychological attitudes which had general implications for adjustment. Social devaluation and feelings of general insecurity were seen as presenting special problems of adjustment. Barker and Wright expected that social workers should be able to alleviate these difficulties by helping the person not to feel inferior and by providing new situations in which he could befrequently successful.

In the following year a revised edition of a bulletin by Barker et al was published. This seminal work in the field of somatopsychology also provided a useful summary of research completed before 1953. In it, the authors reaffirmed the view that physique influences behaviour beyond its influence on the mechanisms of behaviour - in particular physique contributes to the phenomenal characteristics of a person, how he perceives himself and is perceived by others.

By 1953 the view that physical defect would inevitably lead to maladjustment had been somewhat modified. Nevertheless there was still a widespread belief that having the right physique must make personal adjustment easier and life happier - a perpetuation of the classical notion of "mens sana in corpore sano." On this basis Barker et al felt justified in including a discussion of the somatopsychological significance of differences in physical size, strength and attractiveness. In view of the differences we have yet to discuss between cystic fibrosis and visible physically handicapping conditions, the conclusions of this discussion are of relevance to our problem.

Taken together the data suggest that normal variations in physique have relatively little importance for personal or

social adjustment. Barker points out, however, that all of these studies depend for their results on correlational methods and suggests that there are relationships between physique and behaviour which are thereby lost. His critique is indeed a fair one.

A number of independent influences, e.g. sociological, genetic, physiological, psychosomatic and somatopsychological, do enter into these final correlations but there is no reason to believe that these factors will operate to the same degree or even in the same direction. Clearly this is a caveat which will apply in the handling of data in our study. Since physique is only one of many factors determining the behaviour being measured, it may, of course, lose its effect if these other factors are not held constant or experimentally eliminated. Barker makes the additional point that the behaviour measures used tend to be crude and he concludes: "While there is evidence of slight but not negligible relations between measures of physique and personality and social behaviour, methodological shortcomings make it doubtful that this is a true picture of the situation." In view of these shortcomings, Barker gives more weight to the sensitivity of laymen and to the evidence from case studies which suggest that even normal variations in physique can have tremendous influence on behaviour.

Thus, the status of knowledge about the psychology of physical disability, at that time, could be summarised in two sentences:

1. Physically deviant persons are not a homogeneous group psychologically.

2. There is no direct univocal link between physique and behaviour; lawful somatopsychological relationships are mediated by the psychological situation which physique helps to create.

Barker's co-worker Beatrice Wright developed the concepts of the somatopsychological approach still further and her assessment of the field in 1960 adds to our summary. Expanding the first point made above, she pointed out that there was "no substantial indication that persons with an impaired physique should differ, as a group, in their general overall adjustment." She declared that there was more evidence of overlap between the adjustment scores of handicapped and non-handicapped groups than there was of difference, suggesting that most persons with physical defect make as good a personality adjustment to their environment as do non-handicapped individuals.

Furthermore no clear evidence could be cited to show any association between types of disability and personality characteristics.

There are two reservations in this declaration for the liberation of the disabled from the kinds of investigation hitherto reported:-

1. Although personality patterns do not consistently distinguish disability groups, certain behaviours, connected with the particular limitations thereby imposed, do.

2. Although there are no group trends in the evidence, individual studies do show that disability does have a profound effect on a person's life, but not in a direct way nor in a way that is consistent from individual to individual.

This assessment, happily, had the effect of discouraging further studies of this kind which had attempted to correlate the presence of physical impairment with personality or adjustment variables without reference to intervening variables. The question of individual adjustment to disability remains an important one as Professor Tizard (1971) has pointed out: "to the handicapped child and his family, suffering is neither more nor less real whether it occurs in other families or not."

The variables of personality and personal adjustment are still very relevant, then, in a discussion of the implications of physical disability for individuals. We have learned from these early studies that they may best be investigated at the level of individual differences in studies where other important background variables are also taken into consideration.

(b) Studies of attitudes and disability

Until the 1960's the attitudes of people to their own disabilities had been inadequately studied. Systematic studies were few and autobiographical anecdotes abounded. The available evidence suggested a wide variety of attitudes existed which bore more relation to the personality characteristics of the individual than to the nature or degree of his disability. The work of the

somatopsychologists opened up the possibility of discussing the attitudes of the disabled in conjunction with those of their associates in relation to such important practical issues as help, sympathy, curiosity, etc.

The attitudes of the disabled and non-disabled were often found to be in conflict over the meaning of these terms, for example, the help which is offered to a disabled person may be perceived by him as being threatening, because of his fear of social inferiority and dependency. Dembo et al (1956) found their disabled subjects had more bad things to say about being helped than they had good. Wright (1960) provides a most interesting analysis of the grievances and gratifications arising for the disabled in everyday relationships. Two major factors diminish the relevance of this important work for our study. Firstly, these rather subtle social situations are unlikely to cause the same problems for our child patients. Wright is justified in using the argument that the disabled person's management of such social situations will augment or ameliorate the difficulties, to suggest some training in social awareness and skills from an early age. However, the urgency of this matter for this study is further diminished by the second point. It is apparent in this context that the question of visibility is of tremendous importance, for it is the visibility of the handicaps discussed which makes such intrusions into privacy so easy. For these reasons the questions of curiosity, help and sympathy will be discussed only if, and when, they arise in relation to these apparently non-disabled, cystic fibrosis children.

The problem of ridicule and taunting, sanctioned in the adult world, is more likely to arise for our sample. Again,

however, we should prefer to defer discussion of it until occasion demands.

The time has come when we must shift our emphasis once more to consider the attitudes of these 'others' with whom the disabled must deal in the kinds of social encounter described above. The attitudes of others are thought to play an important part in the shaping of the individual's acceptance of his handicap.

The studies reported have assessed attitudes to disabilities and to adults who are thus afflicted. They do not, then, necessarily present an accurate view of the situation for the handicapped child. Quite apart from the concessions and attitude changes made in response to his youth, the child's parents will tend to act as a buffer, protecting him from the harsher aspects of public opinion during his early years. The relevance of studies of social attitudes for this thesis, then, lies in the effect these attitudes have on the child's parents.

As Parsons (1958) has pointed out the meaning of ill health or a disabled physique depends on cultural values. Public verbalised attitudes to the physically deviant have been known for some time to be mildly favourable. An appreciable minority openly express negative attitudes (Mussen & Barker, 1944; Barker et al, 1953.) But, at the same time, indirect evidence suggests that deeper unverbalised attitudes remain hostile (Wright, 1960.)

There are two, not necessarily opposing, approaches which have been put forward to explain the attitudes of the public to disability. The first, a prejudice approach, developed by Wright (1960) and others, is discussed in some detail. The other, invoking a 'body-concept' to account for an individual's attitude to disability, is less well developed and discussed

only briefly.

Typically, attitudes held by the physically normal toward the disabled are held to be negative prejudgments of their personal traits, including what Wright (1960) has called 'devaluation.' She and others see this prejudgment as comparable to the prejudice shown to other minority groups. (Goffman, 1963; Yuker, 1965.) Where normal physique has a high cultural value the disabled are seen as deviant and thereby cast into an inferior status position. By the phenomenon of spread the devaluation tends to extend beyond the limits of the disability and stereotypes evolve. On the basis of these stereotypes all subsequent social encounters are conducted. (Goffman, 1963.)

The evidence available to Wright (1960) did not support this view of a negative stereotype, for publicly expressed attitudes, as we have said, were recorded as mildly favourable or, at worst, indifferent. The suggestion had to be made that other cultural values required that prejudice against the disabled should not be overtly expressed, and that beneath this facade, attitudes were more negative.

Yuker also believed this prejudice toward the disabled to be parallel to that expressed against other minority groups and he and others developed a scale of Attitudes to Disabled Persons (Yuker et al, 1960; Siller and Chipman, 1964; Yuker et al, 1966.) Early work with this scale showed high correlations between scale scores and the amount of contact the individual had with disabled persons, thereby suggesting a means of attitude change. In 1965 Yuker recorded that the person who is prejudiced against the disabled will also tend to believe that all disabled persons are

alike and he was followed in this by Chesler (1965) who related prejudice toward the disabled to ethnocentrism.

It is interesting then to find that Lukoff and Whiteman (1964) who had developed their own scale, would find this view an oversimplification. They could find no evidence of a universal stereotype of the disabled and suggested the need to distinguish between negative attitudes to the condition of disability and negative evaluations of the disabled person, thus arguing against Wright's idea that 'spread' would inextricably bind the two. This distinction between attitudes to disability and to the disabled is to some extent perpetuated in the work relating to the body-concept approach which will be reviewed briefly in a moment.

It is important, before attaching too much weight to these contradictory views of Yuker et al and Lukoff et al to observe that as is so often the case in studies of social attitudes, most of this work is based on studies of university students, the representativeness of whose views must obviously be called into question.

Although it is of interest to know the climate of public opinion of disability, as part of any attempt to understand the plight of the handicapped person and his family, this is not an aspect which will be pursued in our study of cystic fibrosis so that the matter will be left here.

Some of the variables, both of the observer and the observed, which are discussed as determinants of these social attitudes may well be of importance in our study, however. Among the observer variables are sex, age, maturity, level of education and socio-economic status.

Women were found to have more positive attitudes to disabled persons than men. Age, maturity and level of education are thought to be relevant factors but there is a dearth of research evidence. There is no conclusive evidence of social class differences in reactions to disability (Dow, 1965.)

On the other hand, the visibility, cosmetic and functional characteristics of various disabling conditions are often mentioned as important determinants, both of the attitudes of the afflicted and of others.

Attitude studies have most often used the Triandis-Bogardus formulation of social distance to assess the importance of these factors. Siller (1963) and Schontz (1964) suggest that disabilities with least cosmetic and functional implications are reacted to most favourably. Erving Goffman also stresses the importance of these characteristics in his book "Stigma" (1963.) Goffman too argues that physical disability is stigmatising, bringing a change in the attitudes of others in social encounters. The question of the visibility of the handicap is important in relation to the information which is transmitted about the afflicted individual and his condition. Where there is no visible handicap the onus is on the disabled person to decide whether he wishes to be identified as disabled or not. The implications of Goffman's work are important for our understanding of attitudes to cystic fibrosis, a disease which usually lacks visible stigma.

Although there is a paucity of evidence from which to draw firm conclusions about the basis of attitudes to the disabled, some progress has then been made by the view which considers the problem similar to that of studying attitudes to other minority groups.

Some important intervening variables have been highlighted.

Difficulties do still arise for this viewpoint though, in the fact that the prejudgments that are made about the disabled are not always negative. There is still a section of people in our culture which regards suffering and misfortune as enobling and character building in the finest sense of the term. There is no research evidence to support this popular myth.

For the sake of completeness in our small review of social attitudes to the disabled, we should include some mention of the research which has been exploring aspects of the self concept, in particular the 'body concept', as important determinants of attitudes to the disabled. Taken together these studies have provided support for the lobby which seeks to distinguish between attitudes to the physical disability and to the persons who are afflicted by it.

Siller (1963) and Yuker et al concluded that there was a correlation between self image and acceptance of the disabled. Weinstein et al (1964) reported age and sex differences in the value attached to different body parts and it has been suggested that these evaluations might be sources of attitudes to the disabled and to disability. Cormack (1967) used a cognitive dissonance model (after Festinger, 1957) to explore this relationship. On the basis that the degree of discrepancy which exists between real and ideal body concept creates a proportionate degree of cognitive discomfort, Cormack suggested that individuals who experience minimal discomfort in relation to their own bodies would express more positive attitudes to the visibly disabled than those who experienced extreme discomfort. A study of students

offered some support for this view but in general the 'body-concept' approach has very little to contribute to the study to be undertaken here. It is time that some special consideration was given to the more relevant issue of parental attitudes and the parent-child relationship when the child has some physical impairment.

(c) Parent-child relationships

The most significant others in the life of any child are, of course, his parents. It is not surprising then to find a widespread interest in the parent-child relationships of handicapped children dating from the earliest studies in this field.

Allen and Pearson (1928) stressed that the effect of the handicap on the parent-child relationship was one of its most serious consequences, for they saw that relationship as holding the key to the child's future adjustment to the disability, and to his subsequent personality development. Other workers agreed. (Meng, 1938; Kammerer, 1940, and several others.) They showed that the attitudes of parents to their disabled child were more likely to tend to extremes relative to parental attitudes to normal children - oversolicitude, rejection, pressing for achievement beyond the child's capabilities, these were all identified in the early studies. There was some speculation that some of the favourable attitudes and the oversolicitousness exhibited by parents might well be masking deep and inadmissible feelings of hostility to the child. Yugend (1941) felt that parents' own unsolved problems determined their attitudes to this child. Rosenbaum (1943) identified feelings of guilt and responsibility

for the child's condition as the underlying causative factors. Martorana (1954) on the other hand took the opposing view. He compared the personal, emotional and family life adjustment of crippled and normal children and came to the rather original conclusion that it was the children's handicaps that were responsible for the impoverished parent-child relationships and for the child's adjustment. He did not link parental attitude with the child's adjustment to his disability. This rather rare view has been neglected by other workers in this field. Although it is quite possible that Martorana's conclusions are an artefact of his analysis, his thesis does remind us of the importance of remaining open to a multifactorial approach.

Nevertheless, in other studies there is a high probability that the accusation of overprotectiveness will be levelled against the parents of handicapped children at some point, so questions of parental protectiveness and the development of independence should be considered.

It is recognised, of course, that a measure of parental protection is necessary for any child's wellbeing but too much can be harmful. Wright (1960) provides a comprehensive list of 'symptoms' of over protective parents which should guide us in identifying the problem among the parents of cystic fibrosis children. In summary, parents are said to be overprotective when:

1. they are highly child-centred.

   These parents are eager to sacrifice themselves and the rest of the family for one child.

2. they continue to help the child when he is willing and able to help himself.

3. they are inconsistent in their discipline.

4. they are dictatorial and rather arbitrary.

   Accepting parents will give lavish toys and tell the children how to play with them.

   Rejecting parents withhold gifts and privileges or withdraw them on rather slim pretexts.

5. they hang over the child and are given to frequent nagging, criticising.

6. they seek to spare the child every imaginable discomfort.

   Accepting parents do so because they cannot bear to see the child having to suffer.

   Rejecting parents show this anxiety as a cover for more basic feelings of rejection of the child.

7. they will restrict his play.

   Accepting parents fear he will be hurt.

   Rejecting parents are punitive in this way.

8. they deny the child opportunities for growing up.

   Accepting parents seek to keep him as a baby.

   Rejecting parents are unwilling to take trouble to teach him to do things for himself.

9. they do not understand his capabilities and limitations.

10. they monopolise his time.

As an antidote it must be pointed out that this label, overprotective, is in some respects a rather odious value judgment, certainly it requires a fairly fixed reference point of view of what the norm is to be. Cultural and social class differences

might be expected to be influential (Whiting and Child, 1953) and the age of the child in question must also be a major consideration.

The consequences of parental overprotection are usually described in terms of excessive dependency in the child. Again Wright (1960) has catalogued the symptoms of the overprotected child and a precis is given below.

1. He is overdesirous of petting and cuddling. He is often afraid of sleeping alone.
2. He is bossy and aggressive with younger children; oversubmissive and docile with adults and older children. Understandably then, he would probably be well behaved in the classroom but a bully in the playground.
3. He is cheerful when he feels secure, i.e. with his parents, but very inclined to cry and be unhappy when he is separated from them.
4. He shows little or no curiosity.
5. He is overconforming and tends to obey implicitly.
6. He is often apathetic by nature but highly emotional to strong stimuli.
7. He is shy and unfriendly to strangers, not good at playing with other children. Prefers being with parents to being alone. Reads rather than plays with toys.
8. He is jealous of anyone who threatens his position with his loved ones.
9. He is timid and afraid in many situations where there is no real danger.

10. He is pleased by flattery and depressed by adverse criticism. Constantly seeks reassurance and approval.

11. He prefers help in all his activities. Is inclined to reject responsibility.

12. He may be characterised by one of a number of nervous habits which serve as emotional releasers or as attention getters.

It seems that there is a need for caution in decrying dependency or overprotection without considering the broader relationship between the parents and the child. In this summary the scales seem too heavily tipped in one direction although the weight given to ratings of dependency in research reports probably reflects the high value placed on independence in our society. But as long as dependence, conversely, is devalued, independence is distorted as a goal. As Sears et al (1957) point out, when independence becomes the dominating guide of parental behaviour it, too, may lead to emotional insecurity. Shere (1954) found children who were both loved and overprotected to be open, friendly and cheerful in contrast to the behaviour of the children whose parents' overprotection stemmed from rejection.

Thus, although the questions of parental protectiveness and childhood dependency have dominated the literature, and although they are recognised to be significant issues, it is felt that some attempt should be made to restore a better balance in this study. Independence and interdependence go hand in hand and it is important to recognise that dangers exist in excessive demands for independence as well as they do in dependency.

Before considering some ways suggested from the literature to restore a better balance between dependence and independence, it is as well for us to be aware of the parental attitudes said to underlie this behaviour.

A number of parental attitudes have been identified as sources of such overprotective behaviour. Perhaps, surprisingly, genuine love and concern for the child are rarely seen as being the most likely or most important factors. On the other hand, guilt is often mentioned. Parents are reported to feel guilty and responsible for the child's condition, and because of these feelings, may dislike or even reject the child. Certainly the feelings of guilt are thought to drive the parent to indulge the child by way of compensation. This notion is of particular relevance to our study of cystic fibrosis. One could postulate that these parents, having passed on a genetic abnormality, would be particularly susceptible to feelings of guilt and hence to overindulgence and overprotection.

The parents' own need to be needed and wanted is also a possible source of overprotection. Such parents would need to have a dependent child and would be more likely to magnify the child's disability. Another important factor in the parents' own personalities is patience. It takes more patience to allow a child to do something for himself than it does to do it for him, and so-called overprotective mothers may simply be impatient and harassed. These aspects are less amenable to assessment in the course of this study.

No matter what parents do, they will be criticised by someone, and this is particularly true for parents of ill or disabled children, when even more people become involved in

their handling of their child.

Wright (1960) and others suggest some aids to achieving a better balance between independence and dependence such as parents' discussion groups, reading material and opportunities to meet other children. The effectiveness of these media will be examined in relation to the parents of cystic fibrosis children. It seems, however, that much of this literature concerning the parent-child relationship in respect of the handicapped child has become bogged down in a single issue which has never been satisfactorily defined nor described against the reality of the families' home situation. Hewett's (1970) assessment of the status of these concepts of guilt, overprotection and dependency in the literature is a fair one and interesting for its inclusion of the much used terms 'acceptance' and 'adjustment'. Of all of these concepts she says "they describe complex patterns of feeling, attitude and circumstance in terms so grossly oversimplified as to distort almost out of recognition, the original response (part emotional, part rational, part expedient) of the individual parent." Clearly, what is needed is the fresh approach of the most recent studies of individual disorders to be reviewed in a moment. (Hewett, 1970; McMichael, 1971.)

From our review of the research on attitudes to disability a threefold conclusion emerges. There is a need for more systematic investigation of the attitudes of the disabled to their afflictions. To understand the evolution of these attitudes, it is necessary to consider the background of social attitudes, and in particular, the role of the parent-child relationship in the personal development of the handicapped child requires more careful study.

Thus the early literature has paved the way for us now to consider more modern studies of disabled children and striking contrasts are at once apparent. The sterile idealism of the kind of research which sought to measure deviation from norms in isolated characteristics of handicapped children has given place to a more realistic global approach to the problem. At last researchers have begun to acknowledge the need for concern about the many factors, within the child, within his family and within society, that are important in the care of a chronically ill child.

It is rather ironic that it has taken our highly diversified, specialised and sophisticated society so many years of research effort to reach a proposal that was first made many centuries ago: "It is necessary for the physician to provide for the patient himself, and for those beside him and to provide for his outside affairs." Hippocrates: First Aphorism.

(d) Modern Studies: the Global Approach to Chronic Illness

As we said at the beginning, modern medicine has many significant achievements to its credit. Tremendous improvements in preventive and therapeutic measures have been particularly rewarded in paediatric hospitals where mortality rates have dropped spectacularly in the last fifteen years. The price for these improvements has had to be paid in the growing numbers of chronically ill or handicapped children who now survive. As a result, many paediatricians have had to alter their orientation from one of seeking to cure to one of seeking to care for their long-term patients. In the course of this new pattern of medical

care, many problems, of non-organic origin, have arisen and have prompted a change of approach to research in this area.

As we have seen, the early research (pre-1960) had done little to prepare doctors to cope with the barrage of social, emotional, educational and financial difficulties that were to present themselves. From 1960 reports began to appear in the litterature based on the clinical experience of men who had cared for chronically ill or disabled children and had observed the behaviour of them and their families.

Welcome though this return to the global and practical approach indeed was, these were medical men with little or no knowledge of the methods and theories of social science and these intermediate studies tend, if anything, to the opposite extreme of some of the early psychometric investigations. In the latter part of this section we shall see that in the last few years a happy medium seems to have evolved. Teams of clinicians and social scientists are beginning to cooperate to produce more balanced, more carefully constructed accounts of the problems and, more importantly, they are beginning to test possible solutions.

These recent reports of clinical observations and systematic research will be reviewed under the following headings:

(i) clinical experiences of the general problems of chronic illness

(ii) clinical experiences of specific chronic illnesses

(iii) epidemiological surveys

(iv) detailed studies of families having children suffering from particular disorders.

(i) Chronic illness: clinical experiences of the general problems of chronic illness

"In no area of medical concern is this need for a comprehensive approach more discernible than in the management of chronic illness." (Debuskey, 1970.)

Although Apley and MacKeith (1962) were more concerned with a psychosomatic approach to children and their symptoms, they did make room in their book for one of the earliest overviews of the effects of handicap on children and their families. In this chapter their approach became strikingly more pragmatic.

They pointed out that the greatest hazards to the emotional stability of the family of an ill or disabled child lay in maternal fatigue, social isolation of the family and bad management at crisis points. The term 'crisis points' seems something of a misnomer, for the difficult situations which are described do persist over periods of time, but nevertheless Apley and MacKeith's notion of the significance of the adequacy of medical management at these times may well be relevant to the cystic fibrosis study. Five 'crisis points' were suggested.

1. Prediagnostic. If the child has a normal birth and is apparently normal in the first few months there will be growing tension from the moment the first doubt occurs. (Relevant to cystic fibrosis.)
2. Diagnosis. The diagnosis is likely to come as a shock and the way its communication is managed is important but Apley and MacKeith suggest that the

greater crisis of this time comes weeks later when the initial numbness has worn off.

3. School Age. Crises of starting school, potential problems of intellectual assessment.

4. Adolescence. Problems of the possibility of independence and autonomy for the child and of the child's growing awareness of his condition.

5. Parents' Middle Age. Child has become an adult physically but may still be dependent and a drain on the parents. Pressure of responsibility from him may begin to shift to the siblings and cause family tension.

The first of these crises relates to the question of the differences in the problems presented by congenital and acquired handicaps. From the child's point of view Debuskey (1970) suggests that congenital handicaps are more generally accepted by the children though difficulties may arise later because parental attitudes have been recognised or blunt peer group appraisal is manifested. Acquired lesions on the other hand are much more taxing, for the child has experienced his optimal condition and had it taken from him. Kershaw (1966) takes the parental point of view. It is well known that the first question of mothers after giving birth is inevitably "Is he/she all right?" and Kershaw records that the first reaction of parents to a child with a congenital defect is likely to be one of rejection. If the deformity is serious and/or obvious, he claims, there may never be more than an outward show of parental affection. More usually

the desire to atone for feelings of guilt takes over and acceptance of the child, however reluctant, then follows. Kershaw suggests that the congenital abnormalities which are not apparent till rather later are rather similar. He points out that where an initial feeling of rejection has been overcome it may still be lying latent only to reappear when the child has lost his baby attractiveness. In acquired handicaps, unless the previous parent-child relationship was unsatisfactory, disturbances are unlikely to be caused by parental rejection, though excessive protectiveness may be a problem.

It is rather difficult to assign cystic fibrosis to either category on the basis of its being an inherited condition detected at varying intervals of anything from days to years after birth. It seems that, in any case, there would be some difficulty in sorting out the effects of the child's acceptance of his difficulty added to parental problems of adjustment, from the effects of the child's problems of adjustment with lesser parental problems, for the complementary trends might well cancel each other out.

The question of establishing the diagnosis, Apley and MacKeith's second point, is one which Debuskey also sees as critical. His concern extends to the when, where, what and how of communicating the diagnosis to parents and if Apley and MacKeith are right, the management of this situation is important for the family's adjustment to its problems. Debuskey also stresses an immediate corollary to the establishment of the diagnosis, in the prediction of its course and outcome. These factors and their influence will be important in our study. Debuskey is very

sensitive to the problems of adjusting to a condition of grim or, at best, uncertain outlook and differentiates this sharply from the situation where the active disease process has ceased and life is preserved, even though some residual handicap remains. It is this division which has restricted the utility of much of the early research for the study in hand. The situation of the family coping with a child who is blind or deaf or who has an orthopaedic problem or a static kidney defect is surely markedly different from that of the family whose child is still in the grip of an ongoing disease process, however slowly the disease is progressing.

Rather than exert special effort at particular crisis points, Debuskey advocates a pattern of continuous care under which it might be hoped that the last three of Apley and MacKeith's points might be dealt with before they became problematic. This heralds a new era in the care of chronic patients. Organic or physical, emotion, intellectual and socio-economic aspects of the care of the chronically ill child then become centralised. Although different members of the team may contribute to separate aspects of this care, their efforts are coordinated.

In terms of <u>physical management</u> the physician must be willing to consider the needs of the patient and his family in relation to providing treatment. Concessions are not often made to personal factors in the course of physical therapy but Debuskey argues that, in terms of chronic care, this is an important aspect of the situation. For example, it is of great advantage to the family if they can be allowed to become involved in the child's treatment from an early stage to combat the melancholy feelings of helplessness that otherwise set in.

In relation to physical aspects of chronic illness Debuskey (1970) again raises the question of visible versus concealed flaws. Perhaps surprisingly, he finds children with no apparent disability to be more likely to be querulous, withdrawn and irritable than their visibly handicapped peers. Kershaw (1966), on the other hand, stresses the problems for the visibly handicapped and attributes them to the disruptive effect which visible deviance has on the parent-child relationship and on the family's social activities. Although cystic fibrosis does not present any gross deformity, some of the children do have a somewhat abnormal physique the significance of which will be discussed in the research report.

Of the emotional aspects much has already been written. We have seen from the early literature that the emotional responses of the child and his parents are crucial to the child's tolerance and the family's acceptance of the problem. It has been suggested that how the child feels about himself and his problem is a more potent factor in his personal and social adjustment than the nature or degree of his disability (Younghusband, 1970.) It remains for empirical investigation to verify this assertion. From the shortcomings in the literature thus far we have already pressed for these emotional responses of the children to be seen as a function of their age, intelligence and experience as well as simply a reflection of the attitudes of the significant others around them.

This developmental view of the emotional response of children is hardly new but it seems to have escaped the attention of many who have studied sick children. Many aspects

of chronic illness generate fear so it is interesting to note that the literature reports three major sources of fear in children which show a developmental pattern. The most commonly noted cause of distress among young children is the absence of the mother and this separation anxiety is most characteristic of the pre-school age group. Although some degree of overlap is admitted between this and the other two phases there seems to be some dispute over the order of events. Natterson and Knudson (1960) used subjective assessments of fatally ill children from a number of sources to suggest that the dominant fear in the age group 5-10 years was a fear of mutilation which was expressed in the children's reactions to treatment procedures. This phase was less decisive in its expression than the preceding separation anxiety, or than the following fear of death which was thought to be experienced from about the tenth year. Natterson and Knudson suggested the sequence of dominating fears following the same pattern as the maturation of consciousness although their view of this latter was rather original. They held that the child was first aware of his mothern, then of his body and eventually of himself as a finite being, and hence the evolution of his fears.

Debuskey, on the other hand, reports the preoccupation with death in the prepubertal years and sees adolescence, rather, as the time when concern with disfigurement, fear of mutilation, is at its height.

In either event, our study of cystic fibrosis children must take account of this aspect of the children's emotional response. It seems it will be important to understand the

extent to which the child understands what is happening, in order to identify the aspect of the situation to which he is responding.

Debuskey records that some of the behaviour patterns which are generated in defence against these fears may be rather troublesome. Active defences of aggression, be it physical or verbal, may be employed as may the more passive measures of withdrawal and denial. We have already recorded the early speculations that certain disease processes evoke particular emotional ripostes and some writers still hold this view (Younghusband, 1970) but the evidence is not conclusive.

Regarding the emotional reactions of parents, Kershaw (1966) stressed that the parent of a handicapped child is emotionally vulnerable in everything in relation to that child. Certainly Dr. Spock agreed with him for he produced a hefty handbook designed to help parents to deal with the range of problems and corresponding emotions likely to arise in the care of the handicapped child (Spock and Lerrigo, 1965.) Parents are reported to exhibit a gamut of emotional reactions throughout the course of the child's illness. Understandably there is no inevitable pattern in this and marked individual differences in parental response do occur. Nevertheless it is useful to be aware of some of the variations which have previously been recognised in response to certain situations. Initially anger, fear, confusion, shock have been described, giving place to guilt and blame. The discomfort which such feelings cause are said to lead to a search for justification, understanding and meaning in this troublesome situation. Most individual differences are then seen in the reorganisation of the defences

which protect the parents when they have rallied from their early depression. (Hamburg and Adams, 1967). With such turbulence in the parents' reactions it is not surprising that families do alter their attitudes to the ill child. As we have already seen, parental attitudes can vary from being sharply restrictive to being highly indulgent or, worse, can sometimes be seen to vacillate between the two extremes. Debuskey suggests that if some of these changes in parental attitude could be intercepted, the compounding of existing physical damage with unnecessary behaviour distortions might be averted. The study to be reported should take careful note, then, of the emotional responses of the children's parents.

A hitherto neglected aspect, the response of the patient's siblings should be included in discussing the family's emotional response. Akin to the idea that personal suffering is enobling, some hold the belief that siblings benefit from having a handicapped brother or sister (Apley & MacKeith, 1962.) Since the siblings must be involved in the parents' reactions recorded above, risks must exist for these children too. Parents may be less able to give them time and attention. Parental fear of additional misfortune befalling the family may impose unnecessary sanctions on the freedom of the well children too, and the compensatory accentuation of demonstrations of love may be irritating (Debuskey, 1970). Emotional disturbance among the siblings is not then an issue to be overlooked.

Until now we have omitted any discussion of the

intellectual and educational aspects. Although these are questions which must be taken into account in this study, the literature, with its emphasis on special schooling and mental or severe physical handicaps is less relevant to cystic fibrosis. In C.F. there is no evidence of brain involvement and, until proved otherwise, intellectual functioning is assumed to be unimpaired. The physical handicap it poses is not sufficient to interfere with ordinary day to day activities, diminishing the need for special schooling. The particular educational problems of cystic fibrosis children are best discussed in the appropriate section of the research report. Similarly Debuskey's inclusion of socio-economic aspects stresses an area which can only be meaningfully discussed with respect to the prevailing conditions in the family.

(ii) Chronic illness: clinical experiences of specific problems

Although we may later find that the similarities between the problems raised by different handicaps and illnesses are greater than their differences, it is, for the moment, striking that most of the discussions of clinical experience reviewed so far have concerned physical defects of a primarily orthopaedic nature. A few short papers in the anthology edited by Debuskey (1970) help to redress the balance. Of particular interest are the papers concerning the families of children with cardiac complaints, leukaemia and nephrotic syndrome since all of these are internal conditions which are not immediately visible and which do not overtly interfere with the child's ongoing activities. They are thereby apparently more comparable with cystic fibrosis.

Unlike some other authors Debuskey placed his emphasis on the prediction of the course and outcome of the illness and the authors of papers in his book follow his lead.

Heart Disease

Neill discusses the implications of heart disease. In this case the children who survive their first birthday have a chance of being helped to adult life, given good surgical treatment, but 40% of the children thus diagnosed have severe heart defects which make their survival even through the first year unlikely. She finds problems in the predictions of outcomes for parents. In her experience painting too black a prognosis at an early age may create severe adjustment difficulties for the family later if the child survives, and she quotes psychiatrists' reports to indicate the lack of parental acceptance of the child who should have died (on the basis of diagnostic predictions.) The way in which communications are conducted between the family and the doctor are especially important then.

As in the overt physical defects, parents of these children are reported to feel guilt and shame and some react to their feelings by the defence of denial, seen in habitually broken appointments, or in "doctor shopping", i.e. when parents go from doctor to doctor looking for one who will tell them what they want to hear, that nothing is wrong.

Even when these feelings have been alleviated by discussion, the fact that there remains a very real strain in rearing a sick child cannot be denied. Neill is surprised to find broken marriages so rare in her experience of these families

although conflicts are not uncommon and individual parents may seek release in alcoholic oblivion.

As such a child grows older the problems of babyhood give place to the requirements that the child should function adequately at school and make satisfactory peer relationships. Clearly the severity of the child's condition is important in determining his capacity for these things but so too is parental attitude and it is at this phase of life that Neill finds parental over protectiveness most harmful.

Although anxiety and depression are said to be rare among these children guilt and feelings of inadequacy do sometimes occur, especially where there are normal siblings. In summary Neil says, "a cardiac handicap seems to exaggerate and intensify normal behaviour problems and personality distortions, but most who pass through these childhood difficulties resolve them before adulthood."

The problems which he then discusses in relation to adolescence are simply an extension of the normally encountered themes, e.g. ambivalence towards parents, problems of self image and self esteem etc. though made more salient by the illness Neill's conclusion is an optimistic one. With family support he sees the possibility of happy development to the full extent of each individual's capacity as being within the grasp of each of these children.

Leukaemia

The future for the leukaemic child is less bright. Although average survival times are now discussed in years rather than months the maximum is in the order of ten years. In view of

this Sigler (1970) also suggests that there is too much emphasis on the preparation of the family for the early death of the child and not enough attention given to living with it. After all, this is an illness usually marked by physical well being until the terminal stages.

Sigler's approach to the management of his patients is somewhat different from that of other physicians, and it highlights the hitherto neglected point, that the families discussed in such studies as we have reported will not all be in the same situation even objectively. Where there are major differences in the management policy of the attendant physician, the parents' experiences will be different and so reports in this field must be explicit about the background of their subjects.

Sigler acknowledges the emotional upheaval which leukaemia creates for parents but he gives the physician a greater part to play in their support. He does not see the delegation of care to psychiatrists or social workers as being in the parents' interests and does not feel group counselling sessions or parents' group meetings are beneficial. It is not clear from his rationale whether these views express personal prejudice or the special needs of leukaemic parents. Such deviations from the popularly held view are useful, though, in calling into question the sometimes too-readily-accepted conclusions of existing research.

A wide variety of parental defence mechanisms are recognised and seem to be employed in a highly individual and unpredictable way. Sigler sees his main task beyond the medical treatment of the child, as supporting the parents through their emotional problems. He scarcely discusses the feelings of the

feelings of the children although some modified explanation of their condition is given to them. The exact diagnosis and course of the illness are avoided in these discussions.

Nephrotic Syndrome

Sigler's views are in direct opposition to those of Levin who deals with children with kidney problems. Levin (1970) has found it frightening for his child patients if the explanation given to them is inadequate. However the nephrotic syndrome with which he is concerned can be seen and felt by the child himself in his own body as fluid retention makes him swollen and puffy. Most of these children can be treated and have an ultimately good prognosis, although there are unpleasant side effects attached to the drugs used in treatment. Some of the children (20 - 25%) have underlying kidney disease which makes their prognosis much less good, and the problem is that it is not always easy to tell into which category the child falls.

The emotional problems encountered are those which we are finding to be common to serious chronic illnesses; parental anxiety, guilt, confusion, depression creating disturbances in relationships with the medical profession and with the child. Levin calls for early assessment of the resources and deficiencies of the parents in financial, social and emotional terms and on the basis of such an assessment he calls in other members of the team to help.

It has become apparent then from all that has been written, that not only are children with chronic physical ailments more likely to experience psychological and social difficulties, as the early workers suggested, but also the family is likely to suffer stress in any of a number of aspects of family life. Plans are urgently needed for patterns of care which will effectively reduce these secondary handicaps but the personal experiences of clinicians have not entirely solved the problem for us, although they have given invaluable guidelines.

Developments in systematic research in recent times have proceeded in two quite separate directions - to epidemiological surveys and to small scale studies of individual families. We shall discuss examples of each of these developments, briefly.

The epidemiological surveys included are fairly up to date (one of them is still under way), and they give a broad overview of the status of thinking about the problems of chronic physical illness as they are seen at the national level. With this as social background we can then follow the logical extension of the clinical experiences just described, to evaluate the systematic studies of families of children suffering from particular disorders.

(iii) Epidemiological Surveys

Two such studies have been conducted in Britain in recent years. Both have been used to estimate the size and social composition of representative groups of chronically ill children and to present a general picture of some of the social

and psychological consequences. Pless and Douglas in particular have been concerned with estimating the feasibility of introducing remedial measures on a national scale within the existing framework. The argument is that social and psychological malfunctioning occurs among healthy, as well as in chronically sick children, and only to the extent that higher rates of such malfunctioning are found in groups of chronically ill children, in whom the differences cannot be attributed to other variables, e.g. social or demographic factors, can the excess rate of psychosocial handicaps be attributed to the chronic disorder (Pless and Roghmann, 1970).

The Isle of Wight Study (Rutter, Tizard and Whitmore, 1970.) This survey was primarily designed to determine the educational consequences of physical and emotional handicaps. The entire child population of the island, between the ages of nine and eleven, was screened for disability. Children suffering from chronic physical conditions were identified and compared with a small randome sample of healthy children. The incidence of problems in educational achievement and in behaviour was found to be higher than in the control group and, contrary to some earlier suggestions, the overall psychiatric evaluation showed more disturbance among chronically ill children than among their controls. 17% of the chronically ill children, as compared with 7% of their controls, showed psychiatric disorders.

The National Survey of Health and Development.

A representative sample of children born in Scotland, England and Wales during the first week of March 1946 has been studied

at intervals since birth. The vast amount of data thus collected has been put to many uses. Among these, Pless and Douglas (1971) have been examining the psychosocial problems experienced by children who had a chronic illness before the age of fifteen. They have compared their findings with the data on normal healthy children.

Before discussing their results it is necessary to describe their conception of a chronic illness. Chronic illness was defined as a physical, usually non fatal condition which lasted for more than three months of any one year, or which necessitated a period of continuous hospitalisation of more than one month. Pless and Douglas included only those conditions which were of sufficient severity to interfere with the child's ordinary activities to some degree. It is difficult, on the basis of this definition, to see whether cystic fibrosis would be included or not. Certainly it did not appear on the catalogue of illnesses represented in their sample.

Pless and Douglas used three parameters to classify the illnesses included in their survey. They were subdivided by type, motor, sensory or cosmetic; by duration, permanent, indefinite or temporary and by severity mild moderate or severe. Severity was assessed on the basis of the extent to which age and sex appropriate activities of childhood were blocked by the illness. Curiously cystic fibrosis would appear on this system as a mild, permanent illness of indeterminate type.

As in the Isle of Wight Study, educational underachievement was recorded and shown to correlate directly with the severity of the child's condition. It was not clear from the data presented which of several possible intervening

variables mediated in this relationship, e.g. school attendance. Behavioural pathology and maladjustment were again reported and found to be roughly proportional in their extent to the duration of the disorder and only to a lesser degree to its severity. In spite of earlier contentions that it was chronicity per se which made the children vulnerable to psychological sequelae, rather than any specific clinical attribute of their condition, children with sensory disorders were found to be twice as likely to be disturbed as those with physical or cosmetic conditions.

On the basis of these two surveys and a comparable study conducted in the U.S. (Rochester Child Health Survey, Roghmann & Haggerty, 1970), Pless and Roghmann (1971) feel justified in concluding that a high proportion of the social and psychological disturbances recorded must be attributed to the physical disorders. Many of these disturbances are preventable and the notion of comprehensive care has been developed to combat these secondary complications. Although the epidemiological surveys have been useful in verifying these previously familiar, but poorly validated premises, they have not so far produced any useful pointers to solutions. More detailed reports from Pless and Douglas are in preparation.

The group data gathered in such large scale surveys often loses much of the richness of information about the individual case. Thus, although Pless and Roghmann (1971) are able to say that the majority of the children in the study do not succumb to these dangers, they are unable to identify the more subtle predisposing factors. They suggest that, since it is the case that not all chronically ill children exhibit social or psychological disturbance, the allocation of preventive services

on an equal basis for all may not be the most efficient method of coping with the problem.

It seems there is still much to be learned from the study of individual problems at the local level and a number of good comprehensive studies of families of children with particular disorders have been conducted in recent years. Two of these will be referred to here.

(iv) Studies of specific disorders

These two studies provide a most fitting culmination to this literature review, bringing together as they do, all the significant features that have emerged as our understanding of the psychosocial aspects of chronic illness and disability has developed.

Hewett (1970) studied the families of cerebral palsied children. Although the practicalities of dealing with children who have suffered brain damage, leaving them with faulty muscle control and, probably, mental deficiency, are different from those of dealing with cystic fibrosis children, this study does suggest that there is a breadth of common experience among mothers whose children have any kind of chronic incapacity.

Dr. Hewett was concerned to learn of the practical problems these mothers faced and the social services available to help them but, working as she was from Nottingham, she was also concerned to attempt an objective comparison with the family lives of normal children. To this latter end she was able to make use of the celebrated work of the Newsons (1963 : 1968) on the upbringing of normal children in the same geographic region.

Disturbed by the discouraging preoccupations of the literature with the overwhelming problems facing these families, she visited them in their homes to assess the extent of the problems for herself. Having discussed the problems of obtaining the diagnosis, the practical aspects of living with the handicap, its effect on the family, the implications for the child's educational development and the availability and effectiveness of sources of help in any problems that might arise, she was able to record that these families met many of the problems with behaviour that deviated little from that recorded among the parents of normal children.

This is not to deny that there are problems, but simply to point out that the early preoccupation with problems in the literature gives a too biased picture. What Hewett has tried to do is to show in human terms the meaning of these problems for the people most affected by them. It is this sensible approach which has led her to develop the queries we have already noted about use of evaluative terms such as 'overprotective', 'overdependent'. Mrs. Hewett does not presume to advise her readers of where the fine balance point of 'reasonable care of the child' is to be found between the extremes of overprotection and failure to accept the child's handicap. From her data she reports that most mothers do find an optimal position whichbest meets their own and their child's needs, and that, she maintains, is the acid test. Although Hewett dismisses the terms 'acceptance of' and 'adjustment to' handicap as being ill defined and moralistic, to some extent she has tried to explore the feelings, attitudes and circumstances which underlie these concepts.

At times her very practical approach may gloss over real emotional difficulties, but her study has been a useful one in restoring perspective to a previously gloomy and problem-dominated field. It seems it is easily forgotten that often it is the situation in which people find themselves which is abnormal, and that their reaction to it is in fact adaptive, not pathological. As Hewett has said, these parents are required "to accept the unacceptable."

McMichael (1971) conducted a smaller study of older physically handicapped children who attended a special day school where she was the school doctor. Again the predominant disability was cerebral palsy, though other conditions, such as poliomyelitis and haemophilia were represented in her more heterogenous sample.

In some respects her approach was similar to Hewett's. Physical handicap was again described as a hazard and a challenge, that is, the difficulties were not denied nor were they seen as overwhelming; it was seen to be possible for families to surmount the problems; defeat was not inevitable. Being a doctor, McMichael was concerned about the medical aspects of care, and in particular about the translation of the ideal of a team approach to comprehensive care into practice.

Having an older sample, she was concerned with educational problems and includes teachers' assessments of the children among her data. Social problems discussed were in common with those discussed by Hewett, but McMichael gave much more place to the emotional hazards posed by the handicap, not only to the child, but also to his parents and siblings, and she was at some pains to discover the critical factors in their emotional responses.

In this analysis of the families' emotional reactions she was able to bring a fairly objective commonsense approach into an area where assessments have been notoriously unreliable in the past.

McMichael records that many interacting influences bear on the children's ability to make a satisfactory adjustment to their situation, so it is interesting, that from her rather mixed sample of children she concludes that the severity of the child's handicap, and its prognosis, are the two most decisive factors in the child's 'adjustment'. Their influence she sees as multifactorial; "both affect the extent and quality of the mother's anxiety and the element of rejection in that anxiety, and the overall level of stress on the child and his family." (McMichael, 1971.) Parental reactions are clearly important for the child's adjustment then, but so too are the nature and quality of family relationships, particularly the marital relationship. McMichael holds that increased dependency makes the handicapped child even more dependent on these relationships than a normal child.

In the context of the emotionality of handicapped children McMichael also includes an important factor in illness which we have hitherto neglected, the number and duration of the child's hospital admissions. Hospitalisation can be a traumatic experience for any child and the expectation of repeated hospitalisations can have a disastrous effect on the emotional stability of a chronically ill child. We will discuss this again in relation to the data available for the cystic fibrosis children.

McMichael then recognises two sources of emotional difficulty for parents, in anxieties relating to the child but

also in anxieties relating to themselves. In the former category, the severity and prognosis of the child's condition clearly feature, but anxieties about the child's future in terms of education, employment and ultimate care are not overlooked.

In the latter category, very real problems of fears of further pregnancy, marital disharmony, parental ill health and poor social conditions are all seen as contributing to the problem. As Hewett pointed out, there are indeed some families whose resources are already stretched to the limit, and for whom the handicapped child is the provebial last straw.

The main factors affecting the emotional status of the siblings again seem to be the severity and prognosis of the condition and the extent of the anxiety of the mother and of the affected brother or sister.

In seeking to remedy the problems she records, McMichael, like Hewett, sees that some practical changes in the pattern of services and care available could make a considerable difference to the lot of these families.

Both these studies have shown that the primary concern of the early researchers, the influence of physical disability on the personal development of the affected child, is still a most relevant consideration. They have appreciated, as other writers did, that this influence is mediated by a number of factors, principal of which are the attitudes of the child's parents. Although some of the concepts previously used to assess parent-child relationships in this context have been dropped or modified, the unified concern for the child and his family which has emerged is a significant development in this field.

From the literature has emerged a recognition that

clinical insight and objective data are complementary approaches which can fruitfully be combined to give this picture of the child and his family, and that this picture is incomplete without some awareness of other facets of the family's life. Both Hewett and McMichael have taken advantage of these developments in their studies. The points made by the epidemiologists have not been lost sight of, for both authors have shown a realistic concern for the provision of supporting services for the families, not only on a local but also on a national scale. At the same time, the individuality of the children and their families has been maintained, and the importance of the personal significance to them of the problems which arise, has been kept in sharp focus.

It is this balance which we shall attempt to strike in our study of cystic fibrosis children.

Before going on to consider the research which has already been conducted involving these children and their families, it is necessary for us to understand the exact nature of the disease from which the children suffer and the therapeutic measures to which they are thereby subjected.

D. A Layman's Introduction to Cystic Fibrosis (C.F.)

Cystic fibrosis is the most recently recognised of the major chronic diseases of man, having been first described in 1936 by Fanconi in Switzerland. It is still not well known among the general population, nor even throughout the medical profession, although the incidence is thought to lie between 1 and 2 per 2000 live Caucasian births (lower in Negroes and rare in Orientals.) C.F. is now the most common lethal hereditary disease on both sides of the Atlantic, with 400 new cases being diagnosed each year in Britain alone.

This is a particularly interesting chronic disease of children, having a unique combination of factors challenging to the medical profession and relevant to our investigation.

Genetic implications

Cystic fibrosis is known to be an inherited disease although the basic nature of the causal genetic defect is not known. It has a recessive autosomal pattern of inheritance. This means that for a C.F. child to be born, both parents must contribute the defective gene, i.e. both parents are carriers of a gene for C.F. If only one parent contributes this gene the child will become a C.F. carrier but will be phenotypically normal. Male and female children can both be affected; the condition is not sex-linked (i.e., is autosomal.)

The gene is not thought to be isolated in any particular socioeconomic group and cases of cystic fibrosis are found at all socioeconomic levels. It is estimated that between 1/20 and 1/25

of the general population of Britain carries this gene and that around 1/500 marriages in this country take place between carriers. For these couples, both partners being C.F. carriers, there is a 1 in 4 chance for each child they bear, that the child will be affected by cystic fibrosis. Their chances of having an apparently well child are thus 3 in 4. But it must be remembered that a phenotypically well child can be a C.F. carrier as the parents themselves are, thus perpetuating the C.F. gene in the population. Thus the chances of parents who are both C.F. carriers producing a child who is both phenotypically and genotypically free from C.F. are 1 in 4.

As yet there is no reliable screening technique for detection of the heterozygote, i.e. there is no way of identifying C.F. carriers. It is not yet possible to make an intra-uterine diagnosis of C.F. in the foetus.

It is thought that almost all C.F. males are sterile though a few exceptions to this general rule have been reported. C.F. girls, being homozygous with respect to the C.F. gene, have an increased chance of bearing C.F. children. By marrying a carrier the risk of their having a child affected by cystic fibrosis is increased to 1 in 2. Even by marrying a man who does not carry the defective gene all their children will inevitably be C.F. carriers.

Recent recognition

Presumably this gene has passed from generation to generation for some time in the past in this way, but clinical diagnoses of cystic fibrosis have only been proclaimed with

confidence in this country in the last fifteen years. It is surmised that many earlier infantile deaths ascribed to causes such as pneumonia or bowel blockage may well have masked the underlying development of cystic fibrosis in the population. Without exception then, the children in this study are the first generation known to have C.F. in their families.

Paucity of stigmata.

In textbooks (Jolly, 1963) cystic fibrosis is described as a generalised dysfunction of the exocrine glands. In lay terms this means that it affects all the surface secretions of the body, particularly those of the linings of the respiratory tract and pancreatic ducts. The exact nature, distribution and severity of the symptoms can vary widely from individual to individual even within one family. There is no gross visible physical deformity common to the illness, although children whose lung symptoms are severe tend to be barrel-chested, and this can be quite marked in the most severe cases. It is most noticeable when the children are undressed and is fairly well camouflaged by normal clothing. Finger clubbing is associated with pulmonary malfunction and is increasingly exhibited by C.F. children as their lung problems increase. In severe cases it may interfere noticeably with manual dexterity but this is exceptional. On the whole, then, cystic fibrosis children are not visibly deformed.

There is no mental deficiency associated with cystic fibrosis. Because in cystic fibrosis the mucus of the body is altered, the operation of the Eustachian tubes may be impaired

and deafness as a complication of cystic fibrosis has been recorded (Forucci et al, 1972.) Clearly, cystic fibrosis is not contagious then, and it does not pose a serious health hazard to others in contact with the child.

All but the most severely affected C.F. children look, to the outside observer, completely normal.

Problems of Diagnosis.

A few children are identified at birth by a bowel blockage (meconium ileus) which requires surgical intervention in the first days of life. The majority, however, appear as cases of failure to thrive at a some later date, months or even years later. In some cases the symptoms may not be severe enough to attract attention and the diagnosis is missed until some other problem leads to medical investigation.

In some cases, particularly in the past, the symptoms were noticed and medical help was sought but the condition was misdiagnosed. Confusion between the lung symptoms of C.F. and whopping cough or bronchitis, and between the bowel symptoms of coeliac disease or dysentery, were fairly common in the past. In these cases growing anxiety on the part of the mother, the dramatic deterioration of the condition of the child and the exasperation of the local treatment agency they attended, all served to create very stressful circumstances indeed at the time when C.F. was finally diagnosed.

There are three aids to establishing a C.F. diagnosis. The most helpful is the knowledge of a previous diagnosis of

C.F. in the family. The fact that the sweat of these children shows an abnormally high salt concentration has been used in the classical 'sweat test'. Although inherently a good and accurate test it is technically difficult to execute, especially on young infants and corroborative evidence from other sources may be sought before the diagnosis is confirmed. The third test is more complex involving the assessment of enzymic activity in the child's duodenum.

Symptoms : their appearance and treatment.

1. Pancreas

In general this dysfunction of the pancreas means a lack of digestive enzymes in the child's alimentary system which does not allow him to benefit from the food he eats. If the food is not acted upon by these enzymes it cannot be absorbed into the bloodstream for transport around the body. For this reason cystic fibrosis is subsumed under the heading of malabsorptive conditions. Even with treatment it is not possible to achieve completely normal digestion of fat and this will be found in excess in the stools. Excess fat may also interfere with the absorption of food from the intestines. It is not surprising to learn, then, that the untreated C.F. child appears to suffer from malnutrition (Mearns, 1972.) In the classic picture his limbs are usually wasted and stick like, with loose folds of skin covering them. His abdomen is usually grossly distended and in spite of being reported to

have a voracious appetite he shows a poor weight gain. Persistent vomiting, flatulence and bulky, greasy and characteristically foul smelling stools (steatorrhea) also contribute to the picture.

Treatment

(a) A pancreatic extract from hogs or a synthetic substitute may be taken before meals to make good the deficiency in the child's digestive enzymes. This supplement is available in a variety of forms, e.g. pills, capsules, granules, etc. and is not very unpleasant. However, quite a large dosage is required before each meal. The parents regulate the dose themselves guided by the nature of the child's stools, which should approach normal as adequate control is achieved.

(b) Supplementary vitamins are usually prescribed to augment the child's intake in his diet. High levels of vitamin intake are particularly required during periods of fast growth resulting from successful treatment of this disease. The vitamins, available in pill or syrup form, are quite palatable.

(c) A low fat, high protein diet is recommended as being in any case, better for the child's growth but particularly because it is easier for a C.F. child to digest. A fatty diet increases the problem of steatorrhea. Clearly there will be social class differences in the acceptability and, indeed, the financial possibility of providing a suitable diet. Some physicians insist upon an entirely separate special diet for this child composed of high concentrations of nutrients, usually

in liquid form. These are not very palatable or attractive, and the policy in the clinic under study, was to combine a high standard of treatment with maximal care for the child. The advantages of such a diet were not found to be sufficient to justify the unpleasant means.

2. Other symptoms associated with digestive problems.

The children are prone to abdominal pain. It is treatable by medicines given orally. Rectal prolapse may also occur, particularly at the toilet training age. Although it is alarming to see a section of rectum extruding, parents can usually learn to replace it themselves at home and in time the problem ceases. In rare cases, surgical intervention may be necessary. Among the older children additional complications of diabetes and liver dysfunction (cirrhosis) are sometimes reported.

3. Lungs

The mucus lining the respiratory passages normally functions as part of the body's defence mechanism against infection. In cystic fibrosis it is produced in excess and is ineffectively cleared from the lungs. The mucus thus becomes a hazard to the child, blocking the small airways and creating conditions in which infection is encouraged. After repeated infection lung damage becomes irreversible and progressive. It is this aspect of cystic fibrosis which is ultimately threatening to life. During life, the symptoms are an increasingly troublesome

cough, production of purulent sputum and breathlessness on exertion.

Treatment

(a) Physiotherapy (postural drainage) has to be administered several times daily. It is used as a prophylactic measure as well as a treatment procedure to ensure adequate drainage of the lungs. If the mucus is allowed to gather it provides an excellent medium for the growth of infection. Parents are taught how to administer this therapy at home. It takes 10-30 minutes each session and 1-3 sessions a day may be prescribed according to the state of the child's chest.

(b) A variety of aids to physiotherapy have been devised. These, in general, have operated on the principle that if the mucus could be loosened it would be easier to bring it up during physiotherapy. Nebulisers and mist tents have been used. The Nebuliser was a mask held near the child's face so that he would breathe tiny droplets of N. Acetyl Cysteine. The mist tent was erected round the child's bed and he slept enclosed in its moist atmosphere. It was expected that this would loosen the secretions in his lungs before the next morning's physiotherapy. The efficiency of these procedures is now queried. The tent particularly caused material damage to the child's bed, bedding and indeed, sometimes caused problems of damp in the home. The machine required for both the procedures was noisy and unreliable and the aerosol liquid used was expensive. Finally, both procedures were rather unpleasant for the child and inconvenient for the mothers. When attempts were made to assess the benefits

of having such mist therapy, it appeared that the gains were not large enough to justify the effort and this mode of therapy had largely been dropped in Edinburgh. Bisolvon, a medicine to be taken orally, is frequently prescribed now to help the children cough up their sputum (Milner, 1972.)

(c) When infection has set in antibiotics are available to kill different kinds of bacteria, but strains of bacteria can develop which are resistant to the known antibiotics and, in this case, the child's lung infections become a threatening problem. At some clinics constant antibiotic treatment is given with the intention of preventing infection. This is not felt to be good practice in Edinburgh, since it inevitably leads to the establishment of infection with antibiotic-resistant organisms. The children are, therefore, seen very frequently at this clinic so that infections can be diagnosed early. Infections are then treated with a short course of an antibiotic to which the infecting organisms is known to be susceptible. Normally the course of antibiotics can be given at home in tablet or liquid form.

If the child's lungs are not draining adequately and infection has set in, hospital admission may be the only course. In the most severe cases, where the chest infections are caused by a particularly resistant type of bacteria, the only method of treatment is by an antibiotic which can only be given by injection and this, several times per day. This treatment is very distressing for the child, and usually has to be undertaken in hospital, unless the family doctor or district nurse can call frequently at the child's home.

4. Other symptoms

Growth and development are often retarded. The children tend to be small and, particularly the older children, tend to look young for their years. Sexual maturation is achieved thought it is usually delayed. Males, though frequently sterile, are not impotent and they become normal in their secondary sexual development. Females can achieve fertile womanhood.

It has been observed that the poor growth rate in cystic fibrosis (~~in cystic fibrosis~~) can be improved by the administration of anabolic steroids. These drugs have their effect by improving the utilisation of protein. Occasionally they are given to promote growth but more often they are given to those children who have such poor muscle bulk that they are embarrassed by their own thinness. These drugs are not routine treatment, and are given only to selected patients whose problems are particularly severe, since the risk of side effects is high (McCrae, 1973.) Earlier we recorded the high concentration of salt in the sweat as a diagnostic feature of cystic fibrosis. Indeed it is sometimes the first thing mothers notice about their children, that they taste salty to kiss. This loss of salt can be a problem in hot weather when the children sweat profusely and are particularly prone to heat stroke. Salt tablets may be required.

The home treatment programme is then quite strenuous and it is coupled with regular visits to the outpatient clinic for routine examination and tests. Thus treatment can be seen by some families as having as great a nuisance value as the disease itself, and yet, it is only this carefully monitored treatment regime which has brought a future to the children who would

previously have died. Even in the face of these modern developments C.F. is still a fatal disease since the cumulative effect of damage to the lungs may make the situation irretrievable.

To understand how people respond to these particular conditions we turn now to consider the research literature describing cystic fibrosis and their families.

E. Review of the Literature. Part 2: Concomitants of cystic fibrosis

Sociopsychological studies of families affected by cystic fibrosis have only rarely referred to the literature concerning the effects of other diseases and disabilities, and for this reason they do not fall readily into the developmental pattern of approach which was seen to evolve in the review of the literature in general. It may be that other authors have also held Rosenstein's view (1970) of the uniqueness of cystic fibrosis among chronic diseases. Four factors are said to contribute to its special position:

1. Public ignorance. C.F. is not well known when compared with lay awareness of spasticity, leukaemia, etc.
2. Its genetic implications.
3. The tremendous physical involvement required of parents in therapy.
4. Its protracted course and uncertain prognosis.

For all these reasons, and perhaps, too, because of the newness of the problem even to the medical profession, we shall have to evaluate each study, and the experience which it reflects, very carefully in order to extract the fragments of information required to build a coherent picture of the situation of these families.

As in the studies of physical disability already described, much research effort has gone into assessment of the

psychological implications of having cystic fibrosis for the affected child. Although, as we have seen, the consequences of events for parents and children are often inextricably interrelated, for clarity we shall consider first the findings relating to the C.F. children.

Spock and Stedman (1966) elucidated the "psychologic characteristics" of 21 C.F. children ranging in ages from 3 to 16 years, on the basis of scores on the Peabody Picture Vocabulary Test and the Goodenough Draw-a-Person test. Although apparently intellectually brighter, more articulate and more socially adept than average, the children obtained a normal distribution of test scores. Spock and Stedman described the children's verbosity and social conformity as their means of coping with high levels of underlying anxiety. Severe emotional reactions, said to be associated with fear of separation and death, were reported but insufficient data were presented to illustrate this point satisfactorily. In general, the conclusions drawn by this study go too far beyond the evidence which is available to substantiate them.

The study by Lawler et al (1966) was smaller, but more intensive than that of Spock and Stedman. Three separate test batteries were required to present age appropriate tasks to each of the eleven children whose ages ranged from 4-19 years. An intelligence test (Stanford-Binet, the Wechsler Intelligence scale for Children or the Wechsler Adult Intelligence scale), a projective test (the Children's Apperception Test or Rorschach), Draw-a-Person and the Bender Visual-Motor Gestalt tests made up each battery. The children and their parents all underwent

psychiatric assessment.

Major psychological problems were reported in all eleven children while among their parents "marked psychopathology and gross marital discord" were recorded. Again, high levels of anxiety and preoccupations with death were said to be pervasive, and Lawler thus recommends that psychiatric consultation should become an integral part of ongoing care. It is rather curious that, at the same time, he identifies the literature available to these families about C.F. as the major factor in their distress.

Referring to earlier studies of children suffering from other diseases (Dubo, 1960; Glaser et al, 1961; Lynn, 1962), Lawler et al (1966) were concerned with the ways in which the children dealt with their anxiety. On the basis of the psychiatric assessments the children were said to express this underlying anxiety in several ways:

1. Excessive use of denial, leading to complete rejection of treatment.
2. Attempts to obtain good health by magical means, e.g. fantasy.
3. Depression.
4. Hostility to authority figures, e.g. parents, doctors and perhaps teachers, because of the feeling that these adults were allowing them to suffer, thereby letting the children down.
5. Regression.
6. Repressed aggression.

Unfortunately the background information is not available to indicate whence these observations originated, nor are the families' backgrounds discussed, so that these findings can only be tentatively accepted.

More recently, Tropauer et al (1970) have introduced a new slant to these researches by seeking to learn what the children themselves know and understand of their condition. 15/18 children investigated were said to have a fairly good understanding of the disease relative to their intellectual maturity. Few of them expected to be cured and most were aware that the best they could hope for, was satisfactory control of their condition. Even this knowledge in no way ensured cooperation with treatment, and the recurrent danger of sabotage of therapy, particularly by intelligent adolescents, has troubled others who have cared for these children (Pinkerton, 1969; Patterson, 1969; Teicher, 1969; Belmonte, 1969.)

Between 1969-70 several research reports were published which recognised a developmental pattern in the effect of the disease on the children, following the observations of the developmental pattern of predominant fears of children reported by Natterson and Knudson (1960.) Pinkerton (1969) observed that the separation anxieties of young children and fears of suffering and death among older children, are intensified by the children's awareness of their state of health. He warns of the secondary hazards of depression and illness-centredness in these children which may also arise, in response to parental attitudes. It is these difficulties which Pinkerton sees as the source of the troublesome reaction patterns which C.F. children, in his experience, have frequently presented e.g. neurotic hypochondriasis, reactive

depression, sabotage of therapy.

Although interesting, this approach did not offer a means of helping those adolescents who had renounced their treatment, or more directly, those who had attempted suicide, nor did it suggest a course by which the next generation might be guided round this pitfall. Teicher (1969) attempted to extract from his clinical experience of C.F. children, information which would explain how the foundations for these adolescent problems were laid and hence how they might be averted. He points out that for a child to be the object of so much attention, not only medically but also at home and in school, creates a milieu which makes healthy personality development very difficult. Tendencies to poor school work, truancy and a poor work record are summarised by Teicher "they seem unable to follow through on many tasks that demand self discipline." Perhaps this also applies to the therapeutic regime.

Patterson (1969) stresses rather, the socially stigmatising effects of C.F. which are likely to be troublesome in adolescence. Delayed sexual development, dependence on others for help with physiotherapy, the unpleasant cough, even the salty taste when kissed, all these factors, he says, heighten the normal problems of adolescence for C.F. youngsters.

The study by Tropauer et al (1970) was more systematic although clinical observations were also discussed. 23 children aged between 5 and 20 years were interviewed. Their experiences in relation to their illness and its treatment were discussed and the children's fantasies were explored. A modification of the House-Tree-Person technique was used to probe for evidence of psychological conflict.

Even the young children complained of the bother of treatment, e.g. being interrupted at play, dietary restrictions and the physical limitations to keeping up with their peers. Nevertheless Tropauer records that most of the children were quite tolerant of the actual treatment routine, especially when they had had experiences of the consequences of stopping some aspect of treatment for a short time. In such a way the mist tent, for example, could become a symbol of security, in spite of its nuisance value.

Much weight is given to the importance of the children's fantasies and those which are reported from the wishes, dreams and early memories of these young children were said to reflect high anxiety and resentment of the restrictions imposed by cystic fibrosis.

The observations of all these studies of C.F. children are crystallised in the work of McCollum and Gibson (1970.) At the Yale C.F. Service a more thorough investigation has begun, with the aid of 56 families and 65 C.F. children and this is the best of the studies available to date in the literature. So far the only published material comes from interviews with parents and is concerned with the problems arising for the children at different ages. It effectively summarises the most credible aspects of the previous studies.

Children under 4 years of age.

The problems of infancy are said to rest largely on the parents' shoulders.

4-7 years

The child's own anxiety is said to increase during this

time giving rise to inattentive and restless behaviour and frequent daydreaming. Behavioural and disciplinary problems at home and problems of adjustment at school characterise this age group.

8-12 years

This is seen as a time of deepening gloom as awareness of the prognosis emerges at the same time as concepts of death develop. Shame and embarrassment arise from the feelings of difference from the peer group.

Adolescence

All the common problems of this time are further aggravated for the C.F. child. Immature behaviour, excessive dependence on parents, lack of self discipline, avoidance of school and treatment and suicide attempts have been enumerated with a tendency on the part of the children to blame parents and, sometimes the doctors, for everything that goes wrong.

It is now necessary to review the research findings in relation to these parents in order to evaluate this belief. It is perhaps necessary to stress once more that the repercussions of the disease for them cannot logically be separated from the effects which it has on their children, in spite of the efforts of some research workers, so that the separation we have imposed here is a purely artificial one.

Unlike the early trends in the studies of physical disability, the concern with the impact of C.F. on family functioning prceded the study of its psychological effects on

the affected children by two years. Turk (1964) tried to assess the deprivations, financial, social and emotional, which ensued for all family members when cystic fibrosis was diagnosed in the family and she discussed the implications of this situation for family communications.

Her study had several shortcomings. Her sample was rather small (25 families) and rather unrepresentative, being entirely composed of the families of white collar and professional workers. Families with more than one affected child were probably also overrepresented. The testing procedure was very limited, based on a very few forced choice questions augmented by information from some open ended questions which followed.

For all its methodological shortcomings, this study has been much quoted by subsequent researchers for its conclusion that there is a high risk that families affected by cystic fibrosis will experience social isolation and impoverished family communications as a result.

As for C.F. children, the information about C.F. parents is scrappy and in general more emphasis has been given to parents' reactions to the diagnosis than their ability to cope thereafter (Hamburg and Adams, 1967; McCollum and Gibson, 1970.)

McCollum and Gibson remind us that the stress for parents often begins even before the diagnosis of cystic fibrosis. Kulczycki et al (1969) were also interested in the variables which influence parental response to this diagnosis and they stressed the difficulty experienced in obtaining a correct diagnosis and the child's age at the time of the diagnosis as critical factors here.

Difficulties in obtaining the diagnosis may lead to feelings of mistrust and hostility toward the medical profession while the child in its sickly undiagnosed state arouses feelings of self doubt and self reproach in the mother, feelings which may give place to hostility to the child. Clearly, if this state is allowed to persist for long the parent-child relationship may also be damaged.

Even where a definitive diagnosis has been long sought the concensus of experience shows that parents are stunned by this diagnosis and that they seek to minimise the impact of this event by the use of defences, e.g. avoidance or denial (Friedman, 1963; Hamburg and Adams, 1967; McCollum and Gibson, 1970.) As a more realistic view is taken 'anticipatory mourning' reactions are commonly described and the loss or rather the projected loss of the child becomes the central issue. This is described as a time of intense affects and parents at this stage are characterised by somatic complaints, e.g. disturbances of sleep and appetite, by emotional distress, e.g. grief, guilt, anger and by increased motor activity. Intense anxiety is another feature of parents' reactions at this time and this is said to lead them to the next stage of information-seeking and of becoming involved in the minutiae of treatment. These activities are seen as attempts to dissipate their anxiety.

This unambiguous description of parental reactions is not continued to account for parents' long-term adaptation. Only tenuous connections between parental guilt and later overindulgence or, between parental rejection and neglect of treatment, are offered to provide continuity. The factors which

are important are yet to be elucidated, it seems.

Lawler et al (1966) certainly felt there was a need to promote better "psychological adjustment to the disease by patients and their families." He suggested this might best be achieved through the medium of the doctor-parent relationship, a relationship which Pinkerton (1969) felt should be analogous to the parent-child relationship in its compounding of the functions of information giving, support, forbearance and encouragement, with the whole founded upon trust.

It is interesting that the findings of the one systematic study in this area are somewhat at odds with the views of Pinkerton and Lawler et al. Blumenthal (1969) compared the differences between patterns of medical contact experienced by parents of C.F. and of mentally retarded children. She reported some interesting differences in the variety and pattern of professional contacts made, and in the families' expectations of these contacts. Although both groups of families wanted the personal interest of the physician and needed his emotional support it is interesting to note that C.F. parents were said to attach much less importance to obtaining information from the doctors than were the parents of retardates. Some clarification of this point is clearly called for.

For parents the long term burden of caring for a C.F. child is seen as being threefold. (Turk, 1964; Teicher, 1969.) It is financially draining; it is expensive in time and energy and, particularly, it is burdensome to know that, regardless of all the care and effort, the child will never be cured. The toll which this burden takes is variously described as having

deleterious effects on the parents as individuals, on their marital relationship and on their parental relationship, so that the outlook for the family affected by cystic fibrosis would indeed seem to be a gloomy one. Only Tropauer (1969) presents the positive view that in some cases a family may be strengthened and drawn closer by the need to share the problems, although this is an effect that might have been predicted from the work of social psychologists with small human groups.

With, on the whole, such shattering effects predicted for these families, it is perhaps surprising that so few researchers have considered the implications of this situation for the well siblings. Because of its genetic background, cystic fibrosis does indeed make the question of family size a vexed one, but many of the children investigated in the reported research did have unaffected siblings who were ignored in these studies. A little information is available from interviews with mothers.

Tropauer et al (1970) found these siblings very resentful of family sacrifices made for the C.F. child and, because they feel themselves to be deprived in some way, they may well exhibit demanding behaviour and somatic complaints, even feigning C.F., to get attention. Where the family is defensive, avoiding discussion as Turk (1964) described, resentment is often intensified; and the siblings may come to present school adjustment and learning problems and later, delinquent behaviour. Kulczycki (1969) who had previously suggested that C.F. children themselves feel responsible for having cystic fibrosis, considered that healthy siblings might also experience a measure of guilt.

Clearly then the whole pattern of family interaction is worthy of study when one of the children is affected by cystic fibrosis. None of the authors whose work has been reviewed here has yet presented a comprehensive research report embracing the conditions of the whole family. McCollum and Gibson's team at Yale are working on such a project but no further reports have appeared in the literature.

A study of the "Psychosocial Aspects of Cystic Fibrosis" by Denning et al is due to be published in the winter of 1973 but there is little advance information available except to suggest that this, too, may be a more comprehensive study.

At the time when this research was planned there was no such research available and on the basis of this and the many shortcomings of the available studies this project was designed. Before discussing the aims and methods of the present study it is as well for us to be aware of the difficulties these other researchers have encountered.

A number of limitations common to all these studies should be noted:-

1. All of the research literature available was the fruit of work done in the United States or Canada. Quite apart from cross-cultural differences which might reduce the validity of their conclusions for our situation, there is also the question of the fundamental transatlantic differences in the organisation of medical services.

The absence of a National Health Service and the

consequent necessity of paying for treatment impose the new and salient variable of the family's economic situation. We have to bear in mind that a large proportion of the families investigated in these studies were under a financial stress not experienced by their British counterparts.

2. Many of the assessment procedures used have been concerned with obtaining measures of anxiety among parents and children. The results may well be biased by the fact that all the testing to date, even in the McCollum and Gibson (1970) study, has been done in the unnatural and, surely, anxiety-provoking setting of the clinic.

3. Little effort has been made at a global assessment of each family's situation and key family members, e.g. fathers, well siblings are omitted from these studies. As a result some of the relationships between variables by these papers have to be regarded as being very tenuous, often no possible intervening variables were considered. Marital tension is a case in point; where it has been observed it has inevitably been attributed to the existence of the C.F. child, without any discussion of other possibly contributory factors.

4. The very small sample sizes and the lack of control groups further reduce the status of the results reported.

SECTION II The Study

A. Aims of the study

Broadly the aim of this research was to elucidate the social and psychological consequences for families of having children suffering from cystic fibrosis and, in so doing, to make a contribution to our understanding of the concomitants of chronic illness in childhood which would be theoretically interesting, methodologically sound and of practical value while remaining within the constraints imposed by the available resources. From within these lofty ideals a number of more specific aims and sub-goals have evolved.

1. Theoretical Interest.

The study aims to investigate the status of two major variables in the psychology of chronic illness.

(a) Visibility. Cystic fibrosis represents a neglected category among chronic illnesses, being an internal defect which is not visibly disabling. It is hoped that the results of this study may then be usefully contrasted with the findings of the majority of the research reported in the literature, in order to assess the importance of the factor of visibility in studies of physical defects.

(b) Prognosis. The control group of children was selected for the fact that in many practical respects their chronic illness was comparable to cystic fibrosis. There is however a dramatic difference in their prognoses. Cystic fibrosis is still considered to be potentially fatal while coeliac disease, in spite

of the risks it carries of further complications in later life, is not considered to be a life threatening disease. It is hoped that a comparison of the findings from these two groups of families may suggest whether the hitherto somewhat neglected factor of the predicted outcome of a chronic illness has significant influence in psychological investigations of this kind.

2. Methodological Soundness.

Within practical limitations the study aims to meet as nearly as possible those criteria of acceptability already discussed in the critique of the research reported in the literature.

(a) Barker's criteria We have to accept that in multifactorial research of this kind it is virtually impossible to control for, or otherwise manipulate all the relevant variables in such a way as to reveal pure cause-and-effect relationships. At best, we can aim to provide judicious interpretations of such correlative relationships as may emerge. Similarly, the impossibility of achieving a truly representative sample limits the generalisations that can be made from these results. Nevertheless we can aim to redeem some of these problems by the selection of a control group, carefully matched for as many as possible of the variables critical to the account in hand. With such a control group to add perspective to our findings, we can then aim to provide a global account of the life situation of the child suffering from cystic fibrosis, describing his behaviour and personality as carefully as possible, in the light of the severity, as clinically assessed,

of his physical condition.

(b) Criteria imposed by the limitations of previous studies.

In addition to the methodological aims just expressed, the special characteristics of the existing research in cystic fibrosis provide two further sub-goals not included in Barker's schema.

(i) Since this is the first British study of the social and psychological concomitants of cystic fibrosis, it aims to manipulate the economic factor which has proved so significant in the American findings, in a systematic way. To this end, only National Health Service patients and their families are to be included in the sample

(ii) As part of its aim to provide a global assessment, the study sets out to gather information about all family members, from a variety of sources in the families' home environment.

3. Practical Value.

The incidence of cystic fibrosis in the general population is not sufficiently high to make it a defect of common experience. The major aims, and indeed the raison d'être, of this study were thus concerned with the gathering of a body of information which might be of assistance (a) to doctors, in their management of cases of cystic fibrosis, and

(b) to anyone, whether doctor, social worker, teacher or parent,

concerned for the welfare of these children and their families.

(a) To doctors. In its aim to provide information which might be of value to physicians in their management of cases of cystic fibrosis, the study tries to give particular attention to those

issues wherein, practically speaking, there lies most room for improvement. Thus four sub-goals were created:

(i) to examine parents' accounts of their experiences at the time of their children's diagnoses in order to assess whether parents' distress at this time might be alleviated, while their realistic appreciation of the significance of the diagnosis was maintained.

(ii) to assess parents' level of comprehension of their children's disease with particular reference to their understanding of its genetic implications; to detect common misconceptions and to examine the effectiveness of the ways in which information about cystic fibrosis is disseminated.

(iii) to assess parents' attitudes to the system of care into which they have placed their child, and to attempt to identify the aspects of this system to which they are responding. Since parents play a major role in the medical care of their CF children, it is generally held to be vitally important to secure their cooperation and confidence. For this reason this study should aim to include some discussion of parental attitudes to the therapeutic regime, to the organisation of medical services for both out-patient and in-patient care and to the medical personnel who provide that care.

(iv) to record, primarily mothers' experiences in the care of their CF children at home, both in terms of the

children's special medical requirements and of their normal childhood needs; to discuss the usefulness of the services which are available to help these mothers and to point out where needs for assistance remain unfulfilled.

(b) In General. The study aims too, to provide an insight into the concomitants of cystic fibrosis which affect personally, all the members of these families, not only the affected child. Again a number of sub-goals are subsumed under this broad aim:

(i) to describe any practical or social problems which may arise for the family as a consequence of having a child with cystic fibrosis and to search for possible solutions to any problems that may be found.

(ii) to gauge the personal response of parents to this condition in their child and to try to discover those factors which influence their ability to cope. (Within the bounds of this study the concept of parental coping behaviour has a threefold definition. Briefly, the major facets are seen in the parents' ability to look after the sick child; in their ability to fulfil other personal, family and social responsibilities and in their ability to tolerate the situation without disruptive anxiety depression.)

(iii) to assess the influence of cystic fibrosis on the intellectual, social and emotional development of an affected child and to attempt to identify some of the factors through which any such influence may be mediated.

(iv) to consider the implications of this situation for the development of other well children in the family.

(v) to evaluate the uniqueness or more common applicability of these findings, by reference to parallel assessments made of the situation of a carefully matched control group of families.

One further aim runs implicitly throughout this research. This fundamental aim is, that wherever the study may uncover an area of difficulty for these families, it should also try to elucidate the reasons for that difficulty and, where possible, should try to suggest how the situation might be ameliorated.

The theoretical and methodological weaknesses of the study having been acknowledged, it is hoped that they do not erase completely any practical value which the study may have. The author's ultimate aim is to present as interesting, as sound and as valuable a study as possible within these limitations.

B. Design of the Study

1. A note on the Design of this Study.

In the interests of justifying the overall originality of this study and of giving credit to others for particular contributions within it, it is necessary to describe briefly the circumstances of its inception.

The original design formulated during the spring of 1971, was that this study would form one half of a cross cultural comparison of the influence of cystic fibrosis on the lives of affected families in Scotland and Northern Ireland. Information about the Scottish families was to begathered by the author from the patients attending Dr. W.M. McCrae's outpatient clinic in Edinburgh, while information about the Northern Irish families was to be gathered by Dr. L. Burton from the patients known to Dr. J. Dodge in Belfast. Responsibility for this original plan, for obtaining financial support for it and for its ultimate supervision lay with Dr. McCrae.

All four participants then contributed to the construction of the questionnaires to be used in the study. The bulk of the work in the preparation of these questionnaires was borne by Dr. Burton. The author was solely responsible for the selection of tests to be used and for the control study which was conducted only in Scotland. Care will be taken in the presentation of this report to clarify any instances of doubt over responsibilites of this kind.

After this initial period of cooperation the two halves of the project were conducted independently. Shortly after this time too, Dr. Dodge took up employment elsewhere,so that in

practice responsibility for the Northern Irish study lay thereafter entirely in the hands of Dr. Burton.

There had been an initial agreement that any papers which were published or presented at conferences as a result of this work, would appear in the names of all four of the original contributors. 1 paper has been published and 2 others have been read at conferences in recognition of these terms. Two of these three papers were prepared in Scotland.

Unfortunately the agreement was not always maintained. Thus it has been necessary to provide this background information in order to explain why, in a supposedly original work, reference may be found to papers which report the findings of a study almost identical in design to this one, yet which bear neither this author's name nor the names of the two paediatricians. Since satisfactory cooperation could not be achieved it was decided that investigations in the two regions should proceed completely independently.

2. Design of the Study to be Reported.

The study to be reported here gathers data from interviews and psychometric tests in an effort to satisfy the aims avowed at the beginning of this section. Information about the concomitants of cystic fibrosis was gathered first from families visited in their homes in the east of Scotland. The bones of the design of the study may be outlined here and the details filled in by subsequent chapters.

Families were contacted by means of the out-patient clinic or by letter, and appointments thus made to visit them in

their homes. Mothers were interviewed on two separate occasions and fathers were interviewed once, to provide information from two different sources about the impact of the child's illness on family functioning. Factual information, about parents' experiences of their child, his illness and related problems, was gathered at the same time as data about parents' attitudes to the child and to cystic fibrosis. Factors in the parents' personality, which might prove relevant to their ability to cope with the situation, were separately assessed by psychometric test and particular attention was given to the concept of anxiety. The development of young children was assessed by a standardised scale of social competence. The intelligence and attainment of school-age children were assessed in order to judge whether the children were performing to capacity. An independent account of the children's behaviour and performance in school was obtained from their class teachers. The children themselves were interviewed where possible to assess their attitudes to their illness and its effect on their lives. Again, particular attention was given to their anxiety levels which were also gauged by psychometric test. Finally the severity of the child's condition at the time of the study was assessed by the consultant paediatrician.

The design of the study thus allowed interrelationships between behaviour and attitudes and attitudes and behaviour to be discussed, with particular reference to cystic fibrosis.

From this study the significance of the prognosis of a chronic illness, as a factor mediating the effect of that illness on the afflicted child and his family, can be assessed

with reference to a control study of children suffering from another chronic, incurable disorder. Here the control children have coeliac disease. Their complaint is comparable to cystic fibrosis in a number of respects, but strikingly different in that it does not have a fatal prognosis.

The control group of children was then selected to hold constant the influence of the factors of the age and sex of the ill child, and of the socioeconomic status and the size of his family. Interview and test data were gathered as before and compared with the corresponding findings from the C.F. group.

The denouement of this chapter allows us now to consider, in more detail, the nature of these samples and the methods and procedure by which they were described, in the following three chapters.

C. The Samples.

1. Cystic Fibrosis Children and their Families.

By the very nature of the disease the number of families with cystic fibrosis children living in the Edinburgh area was limited from the outset, and an attempt at a 100% sample was deemed the wisest course.

The sample was obtained with the help of the Royal Hospital for Sick Children in Edinburgh, the main centre for the diagnosis and care of cystic fibrosis patients in the region. Current and former patients were included in the sample.

A weekly outpatient clinic is run in this hospital by Dr. McCrae and his staff, to review cases with gastro-intestinal problems. Cystic fibrosis children represent a sizeable proportion of these cases and they have regular, usually monthly, appointments to attend. When closer supervision is called for, the children may be admitted to one of the hospital wards under the care of the same medical staff. Case notes from these two sources provided details of current cystic fibrosis patients.

Not all of the children diagnosed in the hospital remain under its supervision. A few families, in the past have chosen, from the start, to attend a medical centre which is nearer to their homes while other cases are referred from the Royal Hospital for Sick Children, usually because they have become too old to continue as patients of a children's hospital. The hospital's Records Department was able to provide some details

of these former patients and all the surviving cases who were still resident within reasonable travelling distance of Edinburgh, were included in the sample. In the course of this search eight families, whose children were patients of Dr. M. Fraser at the Royal Victoria Hospital in Kirkcaldy, were traced and Dr. Frazer was kind enough to allow us to include them in this study.

It had been hoped that as new diagnoses were made and as families of CF children moved into the region from elsewhere, that these new patients might be incorporated into the ongoing study. Although there were several new diagnoses of cystic fibrosis made during this study, it was decided not to include these families in the sample, for reasons that could not have been foreseen at the beginning of the study.

As the study progressed, evidence was gathered of urgent needs for help, expressed by the parents of these children, which it would have been unethical to ignore. The pointing out of these needs was swiftly followed by an investigation into the ways in which the hospital's pattern of care might be extended, to support such distressed parents in their homes. Thus a home visiting service was introduced and it began where it was most needed, in the follow-up of new patients. It dramatically altered the situation of these families relative to that of their predecessors, but it introduced a problem for this study. It was considered that it would be too much to ask of families that they tolerate the programme of visits required by this study as well as the visits introduced by this new service and that, in any case, this new source of contact between the families and the hospital would

introduce an important, but uncontrolled, source of variance into the families' responses to this study. New patients then, were not included in the sample.

Thus, a sample of 58 CF. children was identified from 50 families living in Central Scotland. The children's ages ranged from 1 yr to 19 yrs 7 m showing a sample mean of 7 yrs. There were 37 boys and 21 girls in the sample, the age and sex distribution of which is shown in Table II(i).

Table II(i) Age and Sex Distribution of the CF Sample.

	Boys	Girls	Total
Preschool Children (5yrs)	18	3	21
Primary School Children (5yrs)	14	14	28
Older Children (11yrs)	5	4	9
Total Number of CF Children	37	21	58
Age Range	1:0yr-19:7yr	4:0yr-17:4yr	1:0yr-19:7yr
Average Age	6:3yrs	8:3yrs	7:0yr

Clearly the age and sex distribution of the sample is not ideal. A preponderance of younger children was to be expected, by the nature of cystic fibrosis, but it is perhaps unfortunate that boys should be so overrepresented, and girls so underrepresented in the preschool age group. This bias in the sample may be important in the interpretation of our findings.

The families ranged in size from having 1 to 9 living children. The mean family size was 2.7. Eight of the fifty families had two children with cystic fibrosis.

For the purposes of socio-economic grouping the classification of occupations (1970) as used by the office of Population Censuses and Surveys, was employed. This system classifies types of economic activity in respect of occupation, industry, employment status and economic position. The families in this sample were assigned to socio-economic groups on the basis of information about fathers' occupations. The socio-economic distribution of the sample, on this scheme, is shown in Table II(ii).

Table II(ii) The Socio-economic Status of the C.F. Sample.

SES Group	No. of Families
I	3
II	8
IIINM	4
III M	30
IV	4
V	-
Services	1
	N = 50

Again the available sample shows a bias which may be important. Since there is no reason to suppose that cystic

fibrosis has an incidence differential between social strata we have to acknowledge that socioeconomic groups IV and V are under represented in our sample.

It is hoped that the significance of these biases in the cystic fibrosis sample may be reduced by the presentation of an adequate control group.

2. The Control Group

There were several reasons for selecting the control sample from children suffering from coeliac disease. A fuller lay account of this condition will be given in Section V but the rationale of this choice of control group may be appreciated without reference to it.

(a) The disease showed a number of similarities to cystic fibrosis:

(i) in its relatively recent recognition by the medical profession and, consequently, in its unfamiliarity to the general public.

(ii) in its being a chronic, incurable internal disorder which thereby presents minimal visual stigma

(iii) in the prediagnostic symptoms and problems which it presents to the family

and (iv) in the responsibility it then places on the parents as the main agents of treatment of their child for an indefinite period of time from his diagnosis

(b) Administratively too, this was an obvious choice of control population. These children attend the same out-patient clinic as the CF children, though somewhat less frequently, and they are cared for by the same medical team. Should in-patient care be necessary, the children would be referred to the same ward.

(c) As we have already indicated there is a striking difference between the two diseases in their outcome. Several of the writers quoted in the review of the literature have ignored the factor of prognosis in their eagerness to point out the commonality of experience among families affected by a variety of chronic disorders. In suggesting that it is chronicity per se, rather than any specific clinical attribute of the particular illness, which is responsible for the psychological sequelae observed, these authors have neglected to discuss the significance of the predicted outcome of the child's condition for the family's situation. The selection of children with coeliac disease for the control study offers an opportunity to make good this omission.

There is one objection to the selection of coeliac disease as a source of the control sample, on the grounds of the difference between the genetic features of the two conditions.

There are a few inherited defects which share the recessive

autosomal pattern of inheritance of cystic fibrosis, most of them are rare, and none of them is comparable to cystic fibrosis in the other salient aspects. Thus objections could be raised on one ground or another to any source of controls that might be envisaged.

The aetiology of coeliac disease is not well understood but there is thought to be a genetic component. The pattern of inheritance is polygenic and the course of its transmission from generation to generation is not yet fully understood. Nevertheless the need for a control group, and the weight of factors for the selection of coeliac disease was allowed to override this objection and, again, appropriate acknowledgment of this must be made in our handling of the data collected.

This genetic issue left a residual problem. Parents who are both carriers of the CF gene face a risk of 1 in 4 that each child they bear will be affected by the disease. On this basis then, it is not surprising to find that eight of the families in the CF sample each had two affected children. Although to have had one child who is a coeliac increases the probability that a subsequent child will also have coeliac disease, the risks are not nearly so high nor, indeed, so well defined. Thus, there are relatively fewer families having two coeliac children. It was too much to expect that such families, living in the Edinburgh region, would match the appropriate CF families in other respects so that the plans for the selection of the control group had to be made with this problem in mind.

The welfare of the children being the major concern, it was decided that their needs could best be examined by the

selection of a control group whose adequacy was judged with first reference to the children's characteristics rather than to those of the family units to which the children belonged. Thus a control group of 58 children was sought to match the C.F. group in the age and sex of the children included, and in the socioeconomic status and size of their families.

The control group was sought by the same means as had been used to locate the C.F. group. Coeliac disease being a much less alarming condition than cystic fibrosis, families thus affected feel much less urgency about their maintenance of contact with the hospitals, and consequently the population was much harder to locate. Clearly, too, there was the problem that many of the available families did not adequately meet the criteria for inclusion in the control sample. Additional patients for the control study were traced, not this time in Kirkcaldy, but in Glasgow. Their inclusion in the study was achieved by kind consent of Prof. D.H. Hutchison and Dr. R.H. Shanks, and with the assistance of the Records Department of the Royal Hospital for Sick Children in Glasgow.

Even with this extended search, it was found impossible to procure adequate controls for four of the cystic fibrosis children. This may in part be due to the slightly unusual nature of each of these cases. Two represented the lower extremity of the age range of the sample; these were year-old twin boys, younger brothers of a four year old child in an SES group III.M family. At the upper limit of the age range no control was traced for the young man of 19 yrs 7 months, middle child in a family of three, in a group III.M household.

Finally, there was a problem in finding a control for a boy of 3 years 7 months whose symptoms were so mild that his parents did not accept the diagnosis of cystic fibrosis and had withdrawn the child from treatment. This child was the youngest of a family of three in a group III.M household.

This situation created a serious problem for the study. Given that the work was felt to be too worthwhile to abandon, and that its value would be drastically reduced without reference to a control group, it was decided that the best available solution was to describe the full cystic fibrosis sample of 50 families and 58 children in its own right, and then to turn to compare the curtailed sample of 54 children with their control group. We have to deplore the necessity for this, and to acknowledge yet another limitation which is thereby imposed on the interpretation of the findings, but when psychology is applied outwith the laboratory then the scientist can do no better than to try to make the best use he can of the existing resources. It is hoped that this idiosyncratic approach may nevertheless be a fruitful one.

Four criteria were applied to the selection of cases for the control group.

(a) Sex The two groups were matched with respect to the sex of the children. Each group then consisted of 54 children, 33 of whom were boys and 21 girls.

(b) Age The ages of the control children were matched to within 6 months of the ages of the corresponding fibrocystic children. Each group then consisted of 18 preschool children (< 5yrs), 28 primary school children (≥ 5 yrs) and 8 older children (≥ 11 yrs). There was no significant difference

between the mean ages of the two groups. Particulars of the age distributions of the two samples are given in Table II(iii).

Table II(iii) The age distribution of the cystic fibrosis group and its control group.

	CF Group	Control Group
Age range	1:2yr-17:4yrs	1:4yr - 17:8yr
Average age	7:1yrs	7:1½ yrs
S.D. (in months)	45	46
	N = 54	N = 54

(c) Family Size The two groups were matched as closely as possible in terms of family size but it was not always possible to procure an exact match. Perhaps because of the nature of cystic fibrosis, and the high risk of its recurrence in a family once affected, there was a tendency for families to be smaller in that group than could be exactly controlled for. The mean size of family in the 54 child CF group was 2.72 as compared with an average family size of 3.05 in the control group. This difference does not reach statistical significance.

(d) Socioeconomic Grouping It was not possible to match all the families on a 1:1 basis from the available population of coeliac controls, and it was necessary to aim only for equivalence in the proportions of each category represented

in each of the two groups. This was successfully achieved and there was no significant difference between the socioeconomic compositions of the two groups. (Table II.(iv)).

Table II.(iv). The Distribution of Socioeconomic groupings in the two samples

	C.F. Group	CONTROL Group
I & II	14	14
III NM	4	3
III M	31	30
IV & V	4	6
Services	1	1
	N = 54	N = 54

(e) A note on the Urban/rural dimension. This dimension is almost a thesis topic in itself. In the absence of a clear-cut system of definition, the umbrella terms 'urban' and 'rural', were omitted from this study, and such concepts were not systematically controlled for in the selection of the control group. This decision is justified by the argument that, a number of important topics e.g. about housing conditions, play facilities, access to medical services, is subsumed under, yet inadequately described by, such terms as urban, suburban or rural. In this study no such assumptions are made and each of the separate features of each family's home environment which is salient, is teased out and considered

individually as a possibly important source of variance in the results.

3. Losses

From 50 CF families there was only 1 father who refused to cooperate in the study under any circumstances, but this was apparently his characteristic attitude to anyone who wished to speak to him, and not a reflection on this study in particular. One mother who had left her husband and children and two fathers, both of whom were living apart from their families, were also unavailable for interview. Thus 49 mothers and 47 fathers of 58 CF children contributed to the study. Two of the children died during the course of the study, but investigations of both families had been completed by the time of their deaths.

On the whole, it was encouraging to hear the enthusiasm and interest which these parents expressed in relation to the study. There was however, rather less enthusiasm about the psychometric testing of parents, where some refusals were encountered. 88% mothers and 82% fathers completed the test battery.

Among the control group, 1 mother refused to participate but an alternative control was found. Ten fathers were missing from the control group of 54 families, although all the mothers responded. The reasons for these losses are tabulated in Table II.(v).

Table II.(v) Reasons for losses of fathers from the control group.

Unmarried mother	1
Dead	1
Separated, divorced	4
Family moving away, father already gone	1
Indirect Refusal (never available)	2
Refusal	1
	N = 10

The enthusiasm for the study among these families was much less than among CF families and they also showed more reluctance to complete the psychometric tests. Ultimately complete profiles were obtained for only 85% mothers and 76% fathers.

D. Methods

1. Parents of chronically ill children : (a) Interviews

There was little difficulty in the choice of the interview as the main method of collecting data for this study. The need to gather information about biographical facts, behaviour and attitudes, from parents of chronically ill children, demanded a specially designed instrument which would satisfy the aims of the study and be acceptable to the respondents. The scheduled interview then, offered an opportunity of gathering the required information in a systematic way while preserving much of the spontaneity and richness which characterises interview data. This method also allowed the investigator to establish rapport with the respondent; to make sure that he understood the purpose of the research in general; and the meaning of each question in it and to ensure that the respondent remained interested and responsive until the end of the interview.

The unrivalled flexibility of the interview was particularly valuable in this study where the existing sample was relatively small in size. Wastage of the sample had to be avoided as far as possible, thereby seriously restricting the opportunities for satisfactory pilot work. Flexibility in the method was useful, therefore, in providing, at least, the possibility of introducing amendments, should experience prove them necessary.

Factors of particular interest which could not be adequately dealt with by an interview, e.g. the child's intelligence, his parents' personality, were assessed by

psychometric tests, the selection of which is discussed later in this chapter.

This study, having been designed to improve upon some of the shortcomings reported in the research literature on cystic fibrosis, was to include information gather from both parents in their own homes. Before interview schedules could be devised, several months of exploratory pilot work were required to crystallise the main topics which were to be covered at these interviews.

It was a relatively simple task to identify the major issues relating to the parents' experience of their child's illness, but it was a more complex matter to identify the factors about which information should be gathered to allow the impact of the illness on family functioning to be assessed. Previous studies of cystic fibrosis were of some help, but as we have said they had their shortcomings in relation to the aims of this study. Hoffman and Lippitt (1960) have drawn up a comprehensive catalogue of variables important in the study of the family life of any child, and their work was useful in highlighting omissions. The work of the Newsons, (1963, 1968, 1970) was particularly instructive at this stage, both in providing methodological clarification as to the way in which major topics in child rearing might be broached with mothers, and in pointing out the significance of the factors of social class and parental attitudes in a study of this kind.

During this exploratory phase of the study, unstructured interviews with key informants and observations of the hospital-based care of the children, modified the list of topics which was

felt worthy of inclusion in the study. The study, at this stage was still part of a cooperative project and Dr. McCrae, Dr. Burton and the author met several times to discuss the pattern of the enquiry. (Dr. Dodge's views were communicated through Dr. Burton.)

A list of topics emerged from these discussions which satisfied all the participants, but which clearly required more information than could reasonably be gathered from one interview with each parent. It was thus decided that the child's mother should be interviewed on two separate occasions, the father once only.

Attention had then to be given to the sequence and wording of the particular questions to be asked, about these topics, the form in which the answers would be required and the way in which these answers would be recorded.

The nature and purpose of the research having been explained to the respondents, it was felt that each interview should begin with sections discussing topics of high face validity and low emotional tone, which would allow a relationship, productive of easy and frank communication, to be gradually developed with the respondent. Discussion of more sensitive issues was deferred until rapport could reasonably be expected to have been established. An attempt was made, nevertheless, to provide logical means of progression for the interviews.

The wording of the questions was not so problematic, as on-the-spot clarification could be provided in individual cases of need. Nevertheless, care was taken to avoid ambiguity, 'leading' and 'loaded' questions.

The approach to the construction of the schedule borrowed from the "eclectic and pragmatic" example of the Newsons (1963), and a variety of question types was used. Overall, the trend was to use simple questions which were amenable to a Yes/No answer or at least to field coding, as a prefix for further open ended questions which then amplified the point to be made. The question of the use of tape recorders in the interviews was raised, but rejected by the clinicians who felt it might not be acceptable to their patients. In retrospect this was a merciful decision from the point of view of the solo investigator; for many of the interviews were long; and the time involved in transcribing from the tapes would seriously have hindered the completion of the study.

Although the principles of the content and design of the interview schedules were discussed at length, the supervision of their preparation was ultimately the responsibility of Dr. Burton. She prepared a draft of the questionnaires which was again discussed and amended, in consultation with Dr. McCrae and the author. Dr. Burton then completed the preparation of the interview schedules for use with parents of cystic fibrosis children and these are reproduced in the Appendix. These shcedules were modified by the author for use in families where two were affected by cystic fibrosis, and later they were modified further for use with the parents of coeliac children in the control group. These latter modifications also appear in the Appendix.

The structure of the interviews conducted in these two groups was naturally almost identical and the essence of the

design of the interview schedules may be appreciated here with reference only to the main topics which guided the nature and form of the questions posed.

In the first interview with parents, usually the mothers of these children, information was gathered under the following major headings, listed in order:

Biographical Information	: this included the age, sex and schooling of the affected child; the age education and occupation of his parents; the duration of their marriage and the total family size.
Housing	: whether rented or owned; adequacy in terms of size and state of repair.
Family and Neighbourhood Support	: frequency and nature of contact with extended family and friends; influence of the child's illness on these social contacts; Help needed, help given.
Contacts with Social Services	: External sources of help - the family's experience of them and their usefulness.

Financial Pressures	: Transport to clinic; extras for the ill child; Sacrifices for the family; holidays; outings.
Illness History	: Events up to the time of the child's diagnosis.
Understanding of the Illness	: The communication of the diagnosis; mother's attitude to the communication and comprehension of it. Discussion of family planning.
Medical Supervision	: Experience of and attitudes to G.P.'s, hospital doctors and outpatient clinics.
Treatment	: Treatment required; child's response to it; mother's problems with it.
Hospitalisation	: The child's experience of and response to being an inpatient. Mother's attitude to hospitalisation.
Communication	: Between family members about child's illness; child's awareness of his illness.
Child's Reactions to Illness	: Age development of the child's reaction to his illness and its symptoms.

Parents' reaction to and Expectations for Child with Illness	: Changes in child rearing attitudes because of illness.
Reactions of Others to Child	: Influence of child's illness on relationship with grandparents, family, friends and siblings.
Developmental History	: Brief account of the child from birth to present including feeding, toilet training, motor and verbal development, general wellbeing, sleep, fears, play and sport; where relevant mother's view of the child's school experience and the attitude of his teachers; mother's view of the child's illness as a handicap.

The second interview for mothers was shorter and more personal. Its main elements are more self explanatory and are listed in sequence below:

Mother's physical and mental health, past and present

Brief family history of illness

Mother's attitude to the child's illness

Domestic pursuits and gainful employment

Mother's expectations re marriage and family

Mother's marital relationship

Her relationship with her other children

Religious beliefs.

Throughout this second interview the recurrent theme is the attempt to assess the influence the child and his illness have had on these aspects of his mother's life.

The interview for fathers compounded these two interviews with mothers. Questions under the following major headings were then, also discussed with fathers:

Father's physical and mental health, past and present

Employment History

Father's understanding of his child's illness

Family Communication

Father's Perception of the Child's Reactions to his Illness

Parents' reactions to and expectations for the ill child

Father's opinion of Handicap

Father's attitude to the child's illness

His expectations re marriage and family

Marital Relationship

Relationship with other children

Religious beliefs

Again an effort was made throughout the interview to obtain fathers' assessments of the influence which their children's illness had had on their lives.

The scale of the study and the severe restriction on the size of the samples which existed did, as we have indicated, make a true pilot study unfeasible. Heavy reliance had therefore to be placed on the clinical experience which guided the development of

the study, and although this clinical acumen was considerable the lack of opportunity for systematic pilot work was felt to be an important drawback at this stage. Particular anxiety existed at this time as to whether parents would be likely to tolerate a research programme which involved a minimum of three visits to their home.

By way of compensation for this initial shortcoming then, the whole study was, in some respects, conducted in the spirit of a pilot investigation, although this was particularly true of the first few interviews conducted. Communication between Edinburgh and Belfast after the first week of the study revealed no major methodological difficulties in the study at that stage. In both groups, parents had expressed enthusiasm for the study and had responded well to the interview schedules. The two investigations then proceeded independently, without major amendment to the schedules, and the data from these first interviews was included in the main study.

One of the problems of the interview as a scientific instrument which we have hitherto neglected to point out, is the question of interviewer bias. In a thesis of this kind it would be a pity if the author had not learned some skills in the course of the study, but, to some extent, the student gains at the expense of the study. Using an instrument of the sensitivity of the interview, the development of the expertise of the investigator during the study must inevitably introduce a source of error to the results. Although the author had had some undergraduate training in the skills of interviewing, an effort was made to improve the base level of performance of the

interviewer before the study was begun, in an attempt to reduce the effect of the interviewer as a source of error. The slow and careful introduction of the investigator to the field of study was also intended to contribute to this end.

Exhaustive though the interview schedules seemed, they offered little opportunity for assessment of the personalities of the parents. The commonly used dimensions of accepting-rejecting, permissive-restrictive, authoritarian-democratic, referred only to the child rearing aspects of the parents' personality. It was felt that the study required more information about personality factors which might be relevant in parents' responses to their child's illness. It was decided then to include psychometric tests in the research programme for parents.

(b) Assessment of parental anxiety levels

The concept of anxiety as a key factor in the response of parents to their child's illness was used repeatedly in the literature. Rarely was any attempt made to define this term, and no criteria were given for the identification of the ubiquitous "high anxiety levels". Within the terms of the study conducted in Edinburgh, an important facet of parental coping behaviour is to be found in their ability to tolerate their situation without disruptive anxiety. Thus it was clearly of value to incorporate in the study some more objective attempt at assessment of parental anxiety levels, even if only in the relative terms of X being more anxious than Y but less anxious than Z.

Among the most important instruments used by psychologists in clinical evaluation are projective techniques. The need to maintain the cooperation of parents in what was already a fairly strenuous research programme was a high priority at this stage, so that the lack of obvious relevance of projective techniques to the study in hand militated against such a selection. This decision was reinforced by the well-known problems of the interpretation of the responses and the quantification of data attendant on the use of projective measures.

There is an objection, too, to the check list approach to the measurement of anxiety, where the subject is asked to identify the statements or adjectives which best describe him. Responses to measures of this kind could be powerfully affected by the vocabulary level and verbal fluency of the subject which might be expected to vary considerably within the unselected samples of this study.

Thus the inventory method of approach was deemed most suitable for the purposes of this study, provided an inventory method could be found which was not contaminated by the effects of linguistic sophistication. The parents would then be asked to assign agreement or disagreement to the items, each of which contributed points to his total score. This method combined quick and easy administration with fairly quick and reliable scoring. Levitt (1968) endorses the view that the advantages of this method outweigh its disadvantages. Nevertheless we must be aware of the difficulties of response set, acquiescence set and social desirability effects to which these methods are

particularly vulnerable. It was hoped that the mode of introduction of the chosen instrument to parents, and the assurances of anonymity might act to reduce these influences.

These decisions narrowed the range of choice considerably, but there remained the selection of the particular inventory to be used, from a large number of scales, most of which appeared to be related to some extent. The use of college students as a population for the construction of scales rendered items in these scales irrelevant to our sample e.g. the S - R Inventory of Anxiousness (Endler et al, 1962). Ultimately the first anxiety inventory to come into general use was chosen - Taylor's Manifest Anxiety Scale (1953).

This scale was one of a number of inventories taken from the Minnesota Multiphasic Personality Inventory. Taylor developed the instrument primarily for use in research to validate the construct of anxiety, but it has been used in clinical settings. Most items in the scale call for a self report of a general condition and words such as 'often', 'hardly ever', 'usually' appear throughout. None of the items requires an estimate of the respondents emotional state at the time of responding; thus we are assessing trait anxiety, or anxiety-proneness, rather than momentary state anxiety. Sample items from the scale may be seen in Table II(vi). A lie scale is included to attempt to overcome the extraneous influences already mentioned, which are inherent in the design of the Inventory.

Table II(vi) Sample items from Taylor's Manifest Scale

I have very few headaches	True/False
I have often felt that I faced so many difficulties I could not overcome them	True/False
I feel anxious about something or someone almost all of the time	True/False

It has been well demonstrated that the scale does not distinguish between normal and psychiatric samples. This instrument also had the advantage of a downward extension of its age range of applicability. A simil r form of anxiety inventory for use with primary school children was prepared by Castaneda, McCandless and Palermo (1956).

(c) Assessment of parental personality factors

It was of interest to this study to be able to describe in more detail the parental personality factors which might influence, significantly, the parents' coping behaviour in this situation. Some estimate of parents' intellectual capabilities was also required, to assist in the interpretation of the data gathered about their comprehension of their child's illness, and their ability to play an active and reliable part in his treatment.

It seemed that both these functions could be fulfilled by the administration of the Cattell's Sixteen Personality Factor Questionnaire (1967-68 Edition) to both parents. One previous objection to this choice was eliminated in 1971 by the

publication of the British Standardisation of this test.

Although on the whole Cattell's work has not invited specific criticisms to the same extent as the work of the other major factor-theorist, Eysenck, it is not without flaws. Vernon (1963) has produced a telling critique of Cattell's factorial conceptions to which we can do nought but concur. He draws attention to the instability of the makeup of factors based on tests,and the lack of evidence for the validity of these tests, hammering home the point with reference to test-taking attitudes and the spurious effects that may result from the response sets to which we have already referred.

In defence we can only submit that the 16 PF seemed the best instrument available to meet the needs of the study and that its weaknesses, having been acknowledged, it should be used with caution.

In fact,some difficulty was experienced in justifying the use of this test to parents. In part, this was a reflection of the attention being given in the public press at that time to personal information being held about individuals, in computer files, and reticence on the part of some parents was readily attributable to this source. Others, justifiably, objected that the time required to complete the 16 PF was too long and others could not be persuaded of its relevance to research concerning their child and his illness. Thus, the 16 PF was included in the research programme for parents, but, neither from a theoretical nor a practical viewpoint was it an ideal choice. The author is inclined to agree with Vernon that the factorial approach is theoretically, unlikely to be - - -"the most fruitful

avenue of progress towards practical assessment and understanding of people's personalities." From a practical point of view it was not perhaps the wisest choice since it did not meet with the approval of all the parents. It remains to be seen from the results whether, from a methodological view, its assessments made any useful contribution to our study.

(d) Note on a discrepancy between the Scottish and North Irish Samples.

On the whole the test procedure designed for use with the Scottish families was adopted in N. Ireland. However one test was introduced in the Irish sample which was not used in Scotland. This was Foulds' Sign-Symptom Inventory. This inventory is of an explicitly psychiatric nature and it was thought that it might not be acceptable to parents. Sample items are quoted in Table II(vii).

Table II(vii) Sample items from Foulds' Sign-Symptom Inventory

Is someone trying to poison you or make you ill in some way?
Do you ever have very strange and peculiar experiences?
Do you have an uneasy feeling if you don't do some things in a certain order or a certain number of times?

Quite apart from the risk of antagonising parents which

the scale represented, it was not felt that it would contribute data which would be useful within the design of the study. In addition, it has to be administered orally and for an 80-item scale the time factor was felt to be prohibitive. The scale was then dropped from the Scottish study.

2. Chronically Ill Children : (a) Intellectual Ability

Although research previously conducted with cystic fibrosis children had been sufficient to endorse the clinical view that the disease does not interfere with mental capabilities, (Spock & Stedman, 1966; Lawler et al; 1966; Kulczycki et al, 1970; Tropauer et al, 1970) some clarification was required on the way in which the children realised their potential. On the one hand it was suggested that the children were more than usually verbal, in response to their high levels of anxiety (Spock and Stedman, 1966), On the other, the severity of the child's clinical condition was thought to play an important part in the underfunction which was recorded by Lawler et al (1966) and Kulczycki et al (1970). It was therefore deemed useful for this study, to assess first the intelligence of these children, and then their educational attainments in order that any evidence of underfunctioning might be brought to light and the factors responsible for it investigated.

Interest having been raised in the question of the children's verbal ability, the Wechsler Intelligence Scale for Children (WISC) was selected for use in this study. This scale provides verbal, Performance and Full Scale I.Q.'s over the age range 5:0 yrs to 15:11 yrs., making it a particularly suitable

choice for this study. Scores are derived from a battery of ten sub-tests, 5 of which assess verbal ability and 5 which contribute to the performance score. The test is well standardised, and modifications of the original American manual and materials have been introduced for use in Britain.

The main drawback of this scale is that it is much more restricted at the extremes of the age range than is Terman-Merrill's revision of the Stanford-Binet Scale. In particular it provides no equivalent to the level of a 4:0 year mental age or below, which is provided by the Binet Scale. This restriction of our intelligence-testing to only the school age children in the sample is not serious, and the choice of the W.I.S.C. is endorsed by Vernon (1962) who finds that - - - -"in most respects this scale (WISC) is preferable to Terman-Merrill."

In accordance with the policy employed throughout the study, of minimising the stressfulness of this study for participants, and, in order to reduce the effects of spurious factors on the test scores, the WISC was given to the children in their own homes. This did introduce the difficulty that in some households testing conditions would not be ideal, but, unless a case of extreme difficulty arose, it was decided to save the children the anxiety of being tested in the clinic setting or the embarrassment of being singled out of a classroom. In most families in fact, every effort was made to provide reasonable test conditions and alternative means of administering the test battery were never required.

(b) Social Competence

Our inability to assess the intelligence of the younger children drew attention to the need to supplement the information already gathered from the interviews with parents, about their development. It was felt that it would be useful to assess, in a standardised way, the rate of progress of the development of the younger children. Although the design of the study would not permit inferences to be drawn from such an assessment to the performance of older children, it was felt that this would, nevertheless be a useful addition to the study. This led to a review of the available scales of social competence.

Social competence has long been a crucial concept in work with children with educational problems, but a paucity of adequate research instruments still exists.

The Manchester Scales of Social Adaptation (Lunzer, 1966) are well developed but they do not cover an appropriate age range for our purposes, beginning as they do at age 6:0 years. The only standardised schedule available in this field was, then, the Vineland Scale of Social Maturity (Doll, 1947; 1965). This is the earliest, best known and most widely used assessment scale of social competence, but it suffers from the disadvantage that its norms are not British.

The scale provides a definite outline of detailed performances in respect to which children show a progressive capacity for looking after themselves. The scale is then scored with the help of information from the mother about the child's achievements in categories labelled self-help, self-direction, locomotion, occupation, communication and social relations.

The child's stage of development is then assessed by the level of performance in these categories which he has achieved. The results may be treated separately to diagnose categories of particular difficulty or they may summed to give a score which is expressed as a Social Quotient. (Method of calculation as for the Binet IQ.)

To some extent, this maturation of social independence must, of course depend on opportunity, a factor for which the scale allows. This factor of opportunity for development is particularly important in the study of these children, whose chronic illness need not be incapacitating, yet whose development might be hindered by the oversolicitousness of their parents.

The criticism that the scale does not provide enough items at each age level applies less to our use of the instrument. There is an average of 11 items in each of the five age levels relevant to our study. This compares favourably with the average of 5 items at higher age levels.

It was thought unlikely that mothers would attempt to boost their children's achievements for the purposes of this scale since the investigator had, by this time, spent several hours in the company of both the mother and child, with the opportunity to observe the child's capabilities for herself.

In spite of its limitations, the Vineland Scale was thus felt to be a useful addition to this study to describe the development of these chronically ill children during their pre-school years (age $\leq$ 5:0 years). It provided an opportunity to assess individual differences both in opportunity and achievement during this time.

(c) Attainment Tests

Returning to the older children ($\geq$ 5:0yrs), and having obtained assessments of their intelligence, it was then necessary to assess their educational attainments. All of the children in the cystic fibrosis sample of this study were attending normal schools at the time of the enquiry, and this simplified the problem somewhat. Nevertheless there are a number of difficulties attendant on educational measurement of the kind envisaged here, and it is as well for us to be aware of the limitations general to this method before considering those of any specific instrument.

It is important first, to be aware of the aspect of the material which is being tested, for children often show unevenness in their performance over different aspects of a subject. By definition, any test of educational attainment reflects not only the child's performance, but also the syllabus and the methods of teaching to which he has been exposed. Geographical and social class factors, as well as differences between individual schools, and between individual teachers are all relevant and combine to detract heavily from the usefulness of national age norms. As well as these factors the familiar sources of error variance involved in any testing procedure cannot be overlooked.

Nevertheless it was important for the study to be able to make some assessment of the achievement of these children, to test whether their illness was indeed depressing their performance. At this stage it seemed appropriate to discuss the requirements of the study with educational psychologists

employed by the Local Authority. The aim was then to select graded tests, which would allow the same test battery to be presented to all the children in the sample, and which would be locally valid.

It was pointed out that, although all aspects of reading do overlap oral pronouncing ability, reading speed and comprehension are relatively distinguishable. It was decided that a test of word pronouncing was required and Schonell's Graded Word Reading Test (1955) was selected. For consistency of approach to the testing of verbal skills, Schonell's Graded Word Spelling Test A (1955) was also used.

A problem was raised by the need to test numerical skills. There have been several revisions of methods of teaching arithmetic and mathematics in recent years and almost all of the available standardised arithmetic tests are now hopelessly out of date. Emphases on the four rules, and on calculations involving weights and measures, have been shaken from their former prominence in primary school curricula, while the advent of decimalisation and metrication has added to the turmoil. The only test which showed any attempt to keep pace with these rapidly changing times was Vernon's Graded Arithmetic - Mathematics Test, for which a decimal currency edition was available (1970). Even this test required calculations in lbs. and ozs., and yards and feet, which would be unfamiliar to some of the children. Caution is therefore required in the interpretation of the results of this test.

One further difficulty was apparent in the fact that the Graded Arithmetic - Mathematics Test was too difficult for the

youngest of the children. It was attempted with all of the children over the age of 6:0 yrs. For the youngest children, and for those over six years of age who could not attempt Vernon's test, skill in arithmetic was assessed solely from the Arithmetic sub-test of the W.I.S.C.

(d) The Goodenough Draw-A-Man Test

Turning from the intellectual development to the personal development of these children, it was of interest to note that drawing tests had been given to cystic fibrosis children as a means of identifying emotional disturbance (Spock and Stedman, 1966; Tropauer et al, 1970). As these tests were primarily designed to assess intellectual maturity, a task in which they are not renowned for their reliability, these interpretations were of some interest to this study. However Harris (1963) published a thorough review of the literature on the interpretation of children's drawings in terms of their creativity, special interests and deep psychological problems or conflicts, and he concluded that there was little confirmed basis for any usage of these tests other than as measures of non-verbal intelligence.

In spite of this, the drawings of cystic fibrosis children have been interpreted in terms of their needs for strength and support or of their feelings of inadequacy. What is interesting though, is the reference to unusual distortions of the representation of the body (Spock and Stedman, 1966). If this were borne out by our study, it was thought that it

might be of interest to the 'body image' school of thought on chronic illness.

Because the test was easy to administer then, and because it was a pleasurable task that might prove a useful 'ice-breaker' in the study of the children, the Goodenough Draw-A-Man Test was included at the beginning of the test battery. It was not intended that the children's drawings should be scored, only that they should be scanned for consistent misrepresentations.

(e) The Children's Manifest Anxiety Scale

As the children were reported to tire easily, and as a good deal of time had already been spent with the family, the testing of the children had to be confined to one session of reasonable length. Thus the only other test that there was time to include in the children's test battery was the junior version of the anxiety scale which had been given to their parents.

Some assessment of the children's anxiety was relevant to the study, both in relation to their health and in relation to their parents' anxiety, and Castaneda et al's version of Taylor's questionnaire (1956) did seem to tap meaningful anxiety manifestations in children. Sample items from this 42 item questionnare are given in Table II(viii).

Table II(viii) Sample items from the Children's Manifest Anxiety Scale

It is hard for me to go to sleep at night	True/False
I wish I could be very far from here	True/False
I worry about what is going to happen	True/False

The questionnaire did include a lie scale to help combat the influence of spurious factors, but its design did have the serious flaw that, the anxiety items scored, all contributed to the total on the basis of an acquiescent response, a particularly serious defect in work with children, who are in any case, usually motivated to please the investigator. Since the test was really only suitable for children over the age of seven years, an attempt was made to combat this shortcoming in the instructions given to the child, but the results must still be interpreted with caution.

(f) Interviews with the Children

With the difficulties of maintaining the interest and attention of the children throughout the testing period, the time factor was critical to this aspect of the study. In order to obtain some information about each child's attitude to his illness, it was decided that an attempt should be made to interview the child. This decision was made at the stage when the research was a cooperative effort, and the preparation of this interview schedule which is included in the Appendix, proceeded as before. Again, the schedule for cystic fibrosis children was ultimately prepared by Dr. Burton, and amended for use with

the control group by the author.

Although a schedule was prepared in the same way as for the interviews with parents, the special problems of interviewing children were acknowledged, and the investigator had a prepared list of topics to raise with the child should a difficulty arise in completing the formal schedule. In fact, with younger children, the author had more success in introducing the topics singly in course of conversation with the child between tests.

An attempt was made to obtain information on the following topics:

School and teachers	-	concerning the child's school activities, friends; Worries in relation to school; His reactions to teachers and theirs to him.
Play, friends	-	Nature and location of play; friends and their response to his illness, the questions they ask.
Ideal person	-	Whom the child would like to be, and why
Family and communication	-	Child's view of his parents and siblings; in whom does he confide.
Illness, Clinic, Hospital	-	the child's understanding of his illness and the treatment it requires; his response to his illness-related experiences.
Feelings	-	What makes the child happy, sad, angry, worried? What is he afraid of?
Fantasies	-	dreams, three wishes, the future.

It was hoped that the information gathered in this way might to some extent make good the necessary omission of other tests e.g. Bene-Anthony Family Relations Test, which might otherwise have usefully been included.

This battery of tests and interviews was intended to describe the development of the child suffering from a chronic illness. It was hoped that it would be sufficient to describe the child, at least as he functioned in the family setting, but the study would not have been complete without some account of the behaviour and performance of the child outside his home, and it was to this end that the last test was included in the study.

(g) The Children at School

In view of the failure of previous researchers to provide information about the behaviour of CF children in natural settings, and their willingness to describe the children as showing varying degrees of 'maladjustment' it behoves this study to test the validity of these descriptions. For this purpose it would be useful to know whether the children exhibit behaviour disturbances in the school setting, and since this is exactly what the Bristol Social Adjustment Guides (Stott, 1970) were designed to do, this instrument was selected for use in the study.

Vernon (1964) aptly describes these guides as a compromise between ratings and more detailed short-term behaviour observations, and as such, they would seem to have

certain advantages over other inventories. There are three separate Guides for the Child in School, the Child in Residential Care and the Child in the Family, of which only the first is relevant to our purposes. This guide offers a number of descriptions of children's behaviour in a school environment, and the child's class teacher is asked to underline those descriptions of behaviour which best fit the child in question. Sample items from the Guide for the Child in School are quoted in Table II(ix).

Table II(ix) Sample items from the Bristol Social Adjustment Guide for the Child in School

	School Work
Manual tasks or free activity:	Seems afraid to begin/difficult to stimulate, Lacks physical energy/ never really gets down to the job and soon switches to something else/invents silly ways of doing things/may spoil his work purposely/ sticks to job.

The items scored are then transferred to a diagnostic chart which potrays "the piling-up of symptoms in particular categories". (Vernon, 1964).

These categories have recently been revised (Stott, 1970)

and are now expressed in the form of five core syndromes:

Unforthcomingness (U)

Withdrawal (W)

Depression (D)

Inconsequence (Q)

and Hostility (H)

Three associated groupings are also recognised in:

Non-syndromic under-reaction

Peer maladaptiveness

and Non-syndromic over-reaction

Scores in these categories are then subsumed under two broad dichotomous groupings of under-reactive and over-reactive behaviour. Scores in these two groupings (and in their constituent categories, if required) can be interpreted with reference to the new norms. (Stott, 1970). This recent reorganisation of the system of classification and the improvements which were thereby wrought in the statistical analysis went a considerable way to answering some of the criticisms levelled at the earlier editions (Vernon, 1964; Chazan, 1970).

In spite of Stott's claims for high inter-rater agreement, reservations were felt about the subjective nature of the report. The standards imposed by different teachers against the background of standards set by the milieu of the particular school were anticipated to introduce new sources of variance with which the design of our study so far could not deal. It was concluded that an additional on-the-spot control

was required for each child in the CF group and for each child in the control group. Thus teachers were asked to select a child from the same class as the child in our study, who most closely satisfied the following criteria:

1. Sex - the child selected was to be of the same sex
2. Age - unless the named child was at an extreme of the class age range a child of about the same age within the class variation were acceptable. More careful matching was requested if the child was unusually young or old for his class.
3. Intelligence - the selected child was to be of equal ability irrespective of differences that might exist between the children's performances.
4. Socioeconomic Status - Teachers were asked to select a child whose family background was of comparable social and economic standing.
5. Family Size and Ordinal Position - Teachers were asked to attempt to match first born, only and late born children. For all other children they were asked only to match at least approximately, the size of families from which the children came.
6. Attendance Record - Where the named child's school attendance was exceptional, teachers were asked to attempt to select a child with a comparable number of absences.

Although it was thought unlikely that all these criteria would be met exactly, the response from teachers was surprisingly good. It was hoped that this introduction of a further control, for the background against which the child's school behaviour was assessed, would be a valuable one.

The Bristol Guide was seen then as a meaningful and useful instrument in the terms of this study. Among its advantages over other rating scales of adjustment, for this study, lay in the fact that it only required the teachers' assessment. Scales developed by Mitchell (Mitchell, 1965; Mitchell and Shepherd, 1966) and Rutter (Rutter and Graham, 1966; Rutter, 1967) had, from our point of view, the disadvantage of requiring a supplementary assessment from parents. However this point was among several which were made in a fairly harsh critique of the Bristol Guides which emerged from the BPS Symposium "Recent Research on Maladjustment" (September, 1967). Although much of the material of this symposium is more properly handled in our discussion, some elements of the critique have to be answered here in order to justify our selection of the Bristol Guide as an instrument for this study.

The criticism of Stott's terms was rather an arbitrary one, and as we have established his classification system has been brought up to date since the Symposium, to include the important concept of inconsequential behaviour (Chazan, 1968). The emphasis in the symposium seems to have been on screening large populations of children for maladjustment, and it was in this context that teachers' assessments alone were said to be invalid for use as indices of maladjustment. This criticism does not seem so applicable to this study where a wealth of information

about the child's behaviour at home had already been gathered from parents, and where all that was required was a description of the child's patterns of behaviour in a different setting.

Sufficient confidence was felt in the usefulness of the Bristol Guide for this same instrument to be applied to the description of the behaviour of the normal siblings of the children in question who were aged between 5:0 and 16:0 years. This was felt to provide a useful supplement to the information available about these children from their parents, and for this purpose the Guides were used as before i.e. teachers were asked to select a child from the same class, to meet the same criteria, to allow a better assessment of any abnormality in the child's behaviour. Since time did not permit a more thorough investigation of the siblings in this study, this assessment was included to probe the advisability of further investigation in this area at some later date.

Although this account completes the battery of materials used in this study it overlooks one difficulty which arises out of the enforced heterogeneity of our sample, that of dealing with the adolescents in the sample. We had already noted from the literature that these young people might be expected to have emotional problems, and that among their problems was the delay in their sexual development which kept them looking younger than their years. With their sensitivity heightened in this way, it was felt that some difficulty might be encountered in obtaining their cooperation in the children's test battery. The upper age limit of the tests used was not then taken as a rigid criterion for their applicability.

After an interview, conducted on the basis of a modified version of the children's schedule, the investigator was able to assess whether the young person would be prepared to cooperate in a testing procedure or not. Those who were not willing and those who were in any case too old for the children's tests, were invited to complete the adult battery of the 16PF and the Taylor Manifest Anxiety Scale. This applied to the young man of 19:7yrs (who was omitted from the control study) and three girls, aged 14:6yrs, 16:2yrs and 17:4yrs. and their controls.

(h) Medical Assessment

Independently of the study conducted by the investigator, two types of assessment were made of the children in the sample. The first was an assessment of the severity of the child's clinical condition at the time of the study.

This was assessed by Dr. McCrae on a three point scale which referred particularly to the children's respiratory symptoms. Thus, children whose physical condition in this respect was good were graded A, the less good B and the most severely affected children were graded C. The investigator was not made aware of these clinical assessments until the study was complete, lest the knowledge bias her approach to the children.

The children had also had routine audiometric tests and the results indicating any hearing impairment were also available to the investigator at the end of the study.

Before considering the fruits of these methods, some clarification is appropriate, of the exact procedure following in the conduct of the study.

E. Procedure

The procedure which was followed in the conduct of this experiment is most clearly described under three major headings:

(1) Preparation for the Study

(2) Conduct of the Study

(3) Treatment of the Results

and we shall now deal with each of these in turn.

1. Preparation

With the additional pressure of the reduced opportunities for pilot work, the need for careful preparation both of the research materials and of the investigator became of paramount importance. It was felt to be necessary that the investigator should fully understand the nature of the disease in question and its treatment before the fieldwork was begun.

The investigator attended regularly the weekly outpatient clinic for cystic fibrosis and coeliac children, sitting in on cases as they consulted, not only the consultant paediatrician, but also the more junior members of his team. This part of the preparation was begun several months before the study commenced and continued throughout the fieldwork. It served several purposes. Besides familiarising the investigator with the nature of the service available to these families and with the kinds of problems which the families present to their doctors, it also served the function of acquainting the families with the investigator. It was quite

customary for there to be an audience in the consulting room so that the author's presence was not unusually obtrusive. It was hoped though, that the parents would not identify the author with the hospital's authority figures, as this could have been a definite obstacle to cooperation in some cases, so that in that respect, there was no positive attempt to be unobtrusive either, i.e. the investigator did not wear a white coat.

Although this familiarisation of the investigator with the operation of out-patient services was very valuable, a deliberate effort was made to avoid discussing particular cases with the staff until the investigation of that family was complete. This was in order to avoid the introduction of bias by the creation of an experimenter expectancy set.

The familiarisation process also extended to the treatment procedures. In the case of the cystic fibrosis children, the hospital pharmacy and physiotherapy departments were able to give practical demonstrations of what treatment involved for them. Coeliac families attending the clinic are advised about their children's dietary requirements by a particularly competent mother who has herself, two coeliac children. This lady was able to give time to explain to the investigator, again with practical demonstration, just what constituted treatment for these children.

In anticipation of problems in handling the data, the author then attended a short course run by the Edinburgh Regional Computing Centre, to learn about the Statistical package for the Social Sciences (SPSS). This provides useful information about the local facilities for the computer

processing of social survey data such as ours.

With the personal aspect of the preparatory work under way there were also some administrative preparations to be made.

As we have indicated,it was felt advisable to consult the Child Guidance Centre of the Local Education Authority in the selection of appropriate attainment tests. When even they had difficulty in recommending an appropriate arithmetic test, the problem was taken to the Godfrey Thomson Unit for Academic Assessment, within the University of Edinburgh,before final selection was made.

Before the Bristol Social Adjustment Guides could be given to teachers to complete, permission had to be obtained from both the Medical Officer of Health and the Director of Education for the City of Edinburgh and for each of the counties covered by the study. Access to the schools having been granted by the authorities, cooperation then depended on the individual headmasters who were approached at intervals during the study.

Where the families included in the study were not Dr. McCrae's patients,it was considered only courteous to advise the families' doctors of the nature of our research. This aspect of the preparatory work was undertaken by Dr. McCrae.

2. Conduct of the Study

In the first instance, the families were approached at the out-patient clinic. It was explained that a study of cystic fibrosis children and their families was in progress, and their participation was invited. A brief account of the nature and purpose of the study was given, explaining that we

were primarily concerned with the problems which the illness posed for the children and their families and with the kinds of help that were required to alleviate these problems. It was stressed that the investigator was in no way medically qualified, that she was seeking to learn from the family not to preach to them. Since it was usually the child's mother who accompanied him to the clinic it was usually possible then to make an appointment to call on her at home for the first interview.

Where families could not be contacted through the clinic, a letter was sent, explaining the study in the same terms and again inviting the family to participate. In this case a provisional time for the first interview was suggested and the onus placed on the mother to reply, only if she did not wish to be interviewed, or if the time suggested was inconvenient. This was found to be much more efficient than having to wait for the mother to confirm an appointment. If there was no objection received from the family the investigator then called at the suggested time and introduced herself.

This two-pronged method of approach was also used in contacting the control group of families.

The first interview could be conducted in $1\frac{1}{2}$-2hrs. but often it took much longer because the mothers became very interested in the topics covered, and wished to enlarge on them. At the end of that interview the structure of the remaining sections of the study was explained to the mothers, and a series of appointments was made to meet the families' convenience and to make optimal use of the investigator's time.

In the normal sequence of events, the mother was next interviewed for a second time. Although this was intended to be a short interview which could be completed in under one hour, again it often took longer not only because of mothers' interest in the questions, but because of their tendency to raise again matters from the first interview over which they had been mulling. From this point of view, contact with the family over time was found to be a very valuable way of obtaining a fuller picture of the family's situation than would have been possible from a single session. At this interview it was explained that we were interested in the factors which influenced parents' responses to this situation, often stressing to the mother that not all mothers coped as she did, and this explanation provided the rationale for introducing the test battery. With assurances that the information so gained was for research purposes only, and that it would be treated with the utmost confidentiality, mothers were then invited to complete the 16 PF and the Taylor Manifest Anxiety Scale. (Mothers were not told that the latter was a scale of anxiety).

If the ill child was under the age of five, the procedure for testing the older children was explained to the mother as part of the rationale for asking her to assist in the completion of the Vineland Scale. These mothers were concerned about the later development of their infants and were encouraged to hear of this aspect of the study.

Where the affected child was at school or older arrangements were made for the investigator to call at a time which suited the child's needs. Although it was possible to see

many of these children in the afternoons when they came home from school, it was felt in some cases that such timing would jeopardise the validity of the findings, since some of the children were reported to be very tired at the end of a school day. Such cases were postponed until Saturday mornings or school holidays.

The testing procedure with the children was normally conducted in the sequence Draw-A-Man, W.I.S.C., Attainment tests, anxiety scale with topics from the interview scattered between the tests. This was found to be most satisfactory with children under the age of about 8 yrs. Older children could attend fairly satisfactorily throughout an interview, and then the schedule was inserted in toto at a point in the test battery selected by the investigator, on the basis of the rapport established with the child.

One difficulty in the testing of these children was the interest that was thereby aroused in the rest of the family. Although siblings could usually be warded off by the investigator, e.g. by asking them to draw things, or by allowing them to play with the W.I.S.C. blocks after the child in question had been tested, disposing of the mother was a more tricky situation. In many cases, the mothers remained in the same room and when they in no way interfered with the test procedure there was no justification for asking them to move. However, the fact that they were within earshot was a serious hindrance to the collection of the interview data. The problem was most acute when the matter of family relationships was raised and children were found to be so inhibited, or to give

such biased answers in this situation, that the question was omitted from the interview or until an opportunity arose to obtain the child's comments in private.

At the end of this testing procedure our desire to obtain information about the child from his class teacher was mentioned to the mother. The way in which this would be gathered was explained and the mother's permission sought.

It was explained to all the mothers who also had healthy children between the ages of 5 and 17 years, that our interest extended to the influence of the affected child's illness on them and that, in addition to their views, we should like to obtain a school report of these children too. Again the procedure for obtaining this information was explained and the mothers' permission sought.

Finally fathers were interviewed. This was often more difficult to arrange, as a number of fathers worked away from home. Sometimes several weeks elapsed between the interview with the father and the study of the rest of his family. although it was usually possible to arrange an evening appointment with the father within a matter of days of seeing his family.

The nature and purpose of the study was explained to the fathers by way of introduction and the interview was con$ducted as described. Again, at the end of this interview, the rationale for including the test battery was explained and the father was asked to complete the tests as before.

The family was then thanked for the assistance it had given to the research project, and assurances were given that feedback would be made available to them at the clinic if they were interested. Indeed, it ultimately became a major part

of the investigator's attendance at the clinic in the later stages of the study to give a progress report to those who had already participated.

When the interviews and tests in the family setting were completed the headmaster(s) of the school(s) attended by the child and his siblings was contacted by telephone. Assurances were given that the release of information from the school had been sanctioned by the authorities, the nature of the study was described very briefly and the part to be played by his school in the investigations was explained. Verbal consent was thus obtained to administer Bristol Social Adjustment Guides to the appropriate teachers. In several cases it was possible to visit the school and discuss the child with his class teacher but in other cases the Guides were sent with a covering letter, which reiterated the basic information of the telephone call, to the Headmaster, who was then responsible for the return of the completed forms to the investigator.

3. Treatment of the Results.

Although the interview schedules had been prepared with computer analysis in mind it was no easy matter to condense the richness of the data gathered into a numerical coding scheme. This was ultimately achieved in a three-stage coding process.

Responses to some questions were immediately amenable to numerical coding e.g. Yes/No items, field coded items of test scores. Other questions had been included in the interview schedule to amplify a single issue of interest. In such cases the separate responses, of less significance in themselves, were

combined and only the cumulated index was coded. Clearly this practice, if carried too far, could involve a substantial loss of information ,so that an intermediate coding process was also used. In this case raw data were coded, as before, on the basis of individual responses but then, later, index values were generated, during the analysis.

Some of the data were in fact analysed manually with the aid of the Olivetti Programma. All of the data were however transferred to punched cards and much of the analysis was done by computer using the Statistical Package for the Social Sciences (S.P.S.S.) The programming of the computer for this analysis was arranged by the User Advisory Service of the Edinburgh Regional Computing Centre, to meet the author's specifications.

The nature and size of the sample,and the nature of the data collected,imposed several restrictions upon the kind of statistical analysis which was appropriate for summarising the results of the study. The mechanical and programming facilities available for the data analysis imposed further restraints,and the outcome was the best available compromise which the existing time and resources would allow.

In a study of this kind the primary requirement was for descriptive statistics i.e. means and standard deviations to express the distributions of the measured variables in a meaningful and convenient form.

Where the closeness of the relationship between two sets of measures of the same individuals was to be described, a coefficient of correlation was required. When the measures in

question were continuous variables, and the relationship between them was approximately rectilinear, then Pearson's product-moment correlation was applied. It is well known that coefficients of correlation take values which are relative to the circumstances under which they are obtained, and that they cannot therefore, be interpreted in any absolute sense. As an aid to the interpretation of the indices of the strength of the relationships thus obtained, between the variables in this study, the statistical significance of each of the obtained correlation coefficients was calculated. In this context then, any coefficient of correlation that is not zero is statistically significant, to the extent of the probability that it is a chance deviation from a population correlation of zero. This interpretation of the Pearson r lifted it from the realm of descriptive statistics to that of inferential statistics, from which the rest of our analysis was derived.

With a great deal of the data expressed in the form of frequencies, in discrete categories, the chi-square test was often a more applicable test of association than such parametric measures as Pearson's r. The constraints on the use of X^2, recommended by Cochran (1954) were observed i.e.

1. In the 2 X 2 contingency table, for $N > 40$, the value of the computed X^2 was corrected for continuity, using Yates' correction, to give a better approximation to the continuous chi-squared function.
2. Again, in the 2 X 2 case when $20 \leqslant N \leqslant 40$ the X^2 test was used only when all the expected frequencies were $\geqslant 5$.

3. The X^2 test was deemed inappropriate for cases where $N < 20$.

4. In contingency tables with d.f. > 1 the criterion for the use of the X^2 test was that no fewer than 20% of the cells should take an expected frequency of less than five, and that no cell should have an expected frequency of less than 1.

In respect of constraint 4 above, it was sometimes necessary to combine categories, and hence to reduce the number of degrees of freedom in order to satisfy these requirements e.g. on the anxiety scale the general population mean score quoted in the literature was 14.5; for the purposes of the X^2 test parents' scores on this scale could be categorised as < 15 i.e. average or below and ≥ 15 above average, although, clearly, care was then required in the interpretation of the results obtained.

When limitation was imposed on the use of the chi-squared test by constraint 3 above, Fisher's exact probability test provided a useful alternative. This test determines whether the two groups in question differ in the proportions with which they fall into the two classifications used.

The second constraint listed did not limit the use of the chi-squared test in the analysis of this data.

In addition to its use as a test of association between variables the chi-squared test can also be used to determine the significance of differences between two groups and in this way it was of value in the analysis, not only of the C.F. group results or the control results in isolation, but also in the comparison of findings from these two sources. In this context too Student's

t test was also valuable. Since the analysis rarely required an assessment of the significance of differences between more than two sets of measurements at a time, a full scale analysis of variance was not really necessary and analysis of the difference between means was usually sufficient. On these occasions the t-test was particularly suitable, being a robust test which is particularly suitable for use in small samples. Although the derivation of the t-distribution assumes that populations from which the samples are drawn are normal and of the same variance, the results of the t-test are not seriously affected if these assumptions are violated. Such robustness is of particular value in a study of this kind where results of two heterogeneous matched samples are to be analysed. The usefulness of the t-test in this study was restricted by the fact that in some cases comparisons were required to be made between findings from groups of differing sizes. In such cases, for example when differing numbers of parents had been available for interview, the results were expressed in percentages and only observable differences were commented upon.

Thus, the statistical treatment of these findings is not sophisticated, nor indeed can it hope to be anymore so than the data which it is to analyse. In this study we have set out to describe both the nature of the variables measured and some relationships between them. The testing of more scientific research hypotheses must wait upon the possibility of a better sampling procedure. Meanwhile the findings reported by this fairly simple but wide ranging study may suggest some such hypotheses worthy of further investigation.

SECTION III - CYSTIC FIBROSIS CHILDREN AND THEIR FAMILIES

The findings of this study will be discussed in four sections. Firstly, the cystic fibrosis children and their families will be considered in their own right. Thus, in this section the interview and test data from this sample are reported and analysed, whilst in the next section, these findings will be interpreted and discussed, with particular reference to the factor of visibility of handicap. Then, the contribution of the coeliac children and their families will be assessed. In Section V, the findings from this control group will be compared with the corresponding data for the matched group of C.F. families. In Section VI, these results will be interpreted and discussed with particular reference to the significance of the factor of the prognosis in the effect of chronic disease in childhood, on children and their families. For clarity, the major topics of this study will be presented in the same order in each of these sections.

In this section, the situation of cystic fibrosis children and their families, as assessed by this study, will be described under the following major headings:

A. The Family Setting

B. The Diagnosis and Treatment of Cystic Fibrosis

C. The Impact of C.F. on Family Functioning

D. The Effect of C.F. on Parents

E. The Influence of C.F. on the Development of Affected Children

F. The Healthy Siblings

A. The Family Setting

In order to obtain an insight into the concomitants of cystic fibrosis, as experienced by these children and their families, it was important for this study first to obtain some background information about the families in its sample, beyond that which was required for the selection of the control group.

The clinical condition of the C.F. children in the sample at the time of the enquiry was thought to be an important background variable in the study. The children were assessed by Dr. McCrae on a three point scale, which referred particularly to their respiratory symptoms. They showed a decreasing incidence of grades from A to C. Table III.A.(i)

Table III.A(i) The Clinical Condition of C.F. Children in the Sample

Grade A - good	47%
B - moderate	31%
C - poor	22%
	N = 58 C.F. Children

Some parental variables also provided an important basis for the study and age, level of educational attainment, occupation and marital status were felt to be relevant in this context. These data, along with information about family composition and housing were gathered from mothers, early in the first interview and are reported in this chapter to depict the family setting against which the subsequent findings should be reviewed.

The mothers of children in this sample ranged in age from 21 to 53 years at the time of the enquiry, showing a mean age of 33 years with a standard deviation of 7.3. Their husbands tended to be older, ranging in age from 21 years to 68 years and showing a mean age of 36 years with a standard deviation of 8.2. These age distributions describe the full complement of 50 mothers, 47 fathers and 3 stepfathers, although not all of them were available for interview.

The duration of parents' formal education was thought to be an important factor, for example, in their ability to comprehend the information given to them about their child's disease, and for this reason the school leaving age of both parents was ascertained. For both mothers and fathers, this lay in the range 14–18 years and, for both groups, the mean age of leaving school was 15.2 years, representing on average, ten years of formal education. In practice, this information was usually supplemented by a rough assessment of the parent's level of intelligence as gauged by his score on Cattell's Factor B, although, as previously explained, test scores were not available for all parents, whereas biographical information was. From the results available from the 44 mothers and 41 fathers, scores on factor B showed a normal distribution of 'sten scores' for both groups.

At the time of the enquiry one third of these mothers was engaged in gainful employment outside the home. The pattern of mothers' employment is shown in Table III.A(ii). The missing case is that of the mother who had left her family. She had not been employed while living at home but she is excluded from this Table, having been absent at the time of the enquiry.

Table III.A(ii). The Employment Status of Mothers at the Time of the Enquiry.

In full-time employment outside the home	4%
In part-time employment outside the home	29%
Part-time work at home	6%
Takes occasional temporary jobs, none at present	4%
No gainful employment	57%
	N = 49 mothers

No corresponding figures were obtainable to describe the employment status of the general population of mothers in the Edinburgh area and the wide age range of the children in this sample made it difficult to make meaningful comparisons with the data provided about the mothers of normal children by the Newsons, although it is of interest that the pattern of employment quoted here is almost identical to that reported for the mothers of normal seven year-olds. (Newsons' data, reported by Hewett, 1970.)

One interesting point does emerge, however, from the exact nature of employment taken by the mothers in this sample and this lies in the rather high proportion of the part-time workers, 8/14, who had taken night-time employment. The significance of this finding may be better appreciated when compared with the pattern of employment shown by the control mothers. It should be added here too, that none of the mothers who had two affected children, of whatever age, were involved in any form of gainful employment.

Fathers' occupations were described by the socio-economic groupings tabulated in Section II, but this does not provide sufficient information for all the purposes of the study. Mothers were then asked whether their husbands had to be away from home at all, beyond the normal working day. Their responses to this question allowed a tabulation of the extent of the fathers' presence in the home, in recognition of both occupational and marital status. (Table III.A(iii))

Table IIIA(iii) Fathers' Presence in the Home

Unemployed, retired	6
Normal working day	25
Shift worker - variable shifts	8
Away from home at irregular intervals	4
Works away from home for long periods	2
Father living apart from family	4
Divorced	1
	N = 50 homes

In view of the genetic implications of cystic fibrosis it was necessary to clarify exactly the nature of the relationships within these families of the 49 women interviewed, all were the natural mothers of the C.F. children in question. However, of the 50 couples who had had C.F. children, only 41 were still together at the time of the enquiry. Two of the mothers had not married the child's father but had married another man. Two of the mothers had divorced the child's father and one

of them had remarried. The remaining five cases represented separated couples. It should also be noted for the interpretation of the findings that three of the mothers in the sample of intact families had previously been married and had had normal children. In these three cases the C.F. children were from the mothers' second marriage.

The duration of the existing marriage was the criterion used in determining how long parents had been married. On this basis, couples in this sample had been married for an average of 11 years. The distribution ranged from marriages of 2 to 32 years and showed a standard deviation of 6.8. It was felt that if the influence of cystic fibrosis on marital stability was to be discussed then the duration of the marriage in question might prove an important consideration.

In none of these marriages was there any kinship, either at the level of first or second cousin, between the partners.

As we have indicated, there are 58 C.F. children in this sample. Three of them were only children but the remaining 55 children had older and younger siblings, bringing the mean family size, for the sample as a whole, to 2.72. However this figure refers only to the surviving children. Ten of these fifty families had lost children before the study was begun, indeed in one family had lost three, and another, two children, so that the total number of children who had died was 13. Information about the family composition of these families is given in Table III.A(iv.)

Table III.A(iv) Family Size and Composition of C.F. Families

Total No. of Families	= 50
No. of families having 1 C.F. child	= 42
No. of families having 2 C.F. children	= 8
No. of C.F. children	= 58
No. of healthy siblings	= 78
Mean family size (living children)	= 2.72
No. of deaths	= 13
No. of miscarriages	= 25
No. of terminations	= 7
Mean No. of conceptions per family	= 3.6

It may well be significant for the implications of this mortality rate that in eight of those ten families, the deaths recorded represented babies, usually diagnosed as having had pneumonia. It is only with hindsight that we may postulate that these infants might well have had cystic fibrosis. In one of the remaining families, the child who died was three years old and diagnosed only at post-mortem. Thus the experience, among these families, of the death of children known to be suffering from cystic fibrosis is confined to one family in which two children died at the ages of 1 year and 6 months, and is thus considerably less than the mortality rate might imply.

More than one third, 34%, of these 50 women also reported miscarriages. Four of them had miscarried twice and two, thrice.

Six of the fifty mothers had undergone a termination of pregnancy and indeed one of them had done so twice. Thus the actual family size underrepresents the total number of conceptions in many families. Had all pregnancies been allowed to go to term and had all the children survived, the mean family size would have been 3.6.

Finally in this chapter, we need to describe the home background of these children and their families. Three aspects of their housing conditions were thought to be of importance for this study: overcrowding; adequacy of the state of repair and facilities, and the effect of having a child with cystic fibrosis in the house.

Following Douglas and Blomfield (1958), a crowding index was arrived at by taking all members of the household, including children, and dividing their number by the total number of occupied rooms, excluding the kitchen, unless it was also used as a living room. Overcrowding was described by a density of more than $1\frac{1}{2}$ persons to a room. On this basis 16% of families were said to be living in overcrowded conditions. One of these was a temporary condition, while another two families were awaiting removal to another house, so that overcrowding not constitute a common problem of housing among the families in this sample.

24% of the families in the sample owned their own homes. Two were living temporarily with their families at the time of the study and one house was provided with the father's job. The remaining 70% of the families lived in rented accommodation. The adequacy of the state of repair and the facilities of these homes was assessed by the investigator by observation only. The scale

by which the adequacy of the housing was assessed is shown in Q27 of the first interview schedule and in Table III.A(v).

Table III.A(v) Index of Housing Adequacy

	3	2	1
Exterior repair	good	moderate	poor
Interior repair	good	moderate	poor
Interior comfort	good	moderate	poor
Garden facilities	good	moderate	poor
	Index takes values 00-12.		

A poor state of repair, whether interior or exterior, described homes which were only questionably fit for human habitation, showing obvious signs of damp or disintegration. Moderate repair described homes of which the fabric required some attention but which were still in reasonably habitable condition. Good repair, described houses which appeared structurally sound. Comfort was assessed in a similar way with reference to the basic furnishings required by the family for living, eating and sleeping. The source of warmth for the inhabitants was also considered. Conditions meeting the families' basic needs were described as moderate. Where these needs were not met, conditions were said to be poor and where they were exceeded, conditions were said to be good. Garden facilities were assessed by the availability of a fenced-in piece of land in which a child could play in safety

within easy access of his home and his mother. Thus multi-storey flats, however opulent inside, scored 'poor' in this respect. Any private reasonable sized piece of private garden scored good and shared plots, tiny yards and patios scored moderate. The question of play facilities included in Q27 was not equally relevant to all the families in the sample so it was not included in the housing index.

The families in the sample obtained scores on this index which ranged from 4 to 12, showing a mean index value of 9.4 with a standard deviation of 2.0. The investigator's assessment showed a significant degree of concordance with the mothers' views about the adequacy of their homes ($X^2 = 14.41$, d.f. = 1, $P < 0.001$)

In the past, structural damage to houses had been reported due to the use of the mist tent. Only 12% of the families in this sample had experienced difficulties of this nature. Although in a few cases the dampness had damaged furnishings it had rarely caused structural damage and in all of these cases the use of the tent had been discontinued.

Local authorities do not have cystic fibrosis on their list of medical conditions warranting note in the Housing Department so the C.F. families obtained council houses, and had them maintained, through the usual channels. Concessions had been granted to two families, under different local authorities, on the basis of the child's condition, and both these families were thus able to obtain ground floor houses with gardens, rather than the flats to which they would otherwise have been allocated.

Whatever the merits or shortcomings of the families'

accommodation the wider question of the neighbourhood in which the family lived had also to be considered. It may be important for the interpretation of later findings to know that all but eight of the fifty families had been living in their present homes for at least one year by the time of the study. Only three of the mothers actively disliked the neighbourhood they lived in, although a further thirteen of the fifty mothers had some reservations about it. Thus ~~only~~ 64% of the mothers said they liked the neighbourhood in which their homes were situated.

The brief insight provided by this chapter into the background of these families, will be amplified in Part C of this Section. An introduction was required at this stage, however, to give depth to our understanding of the findings describing the diagnosis and treatment of the children in these families.

B. The Diagnosis and Treatment of Cystic Fibrosis

The attitudes of these parents to the diagnosis and subsequent care of their cystic fibrosis children were intrinsically interesting, for they offered insights into ways in which the medical management of such cases might be improved. These attitudes were also the sine qua non of the second focus of our study, the influence of cystic fibrosis on the family, for it was through these attitudes that much of that influence was mediated.

Parents' experiences from the time of their child's diagnosis to the time of the enquiry were carefully documented and it is these findings which are presented first, in order that the subsequent results, relating to the myriad repercussions of cystic fibrosis, may be appreciated in proper context. Parents' attitudes and experiences are recorded here under six main headings:

1) The Diagnosis of Cystic Fibrosis
2) Parental Comprehension and the Genetic Implications of Cystic Fibrosis
3) Medical Supervision
4) Treatment
5) Agents of Help
6) Hospitalisation

The questions from which this information is derived may be found on pages 9 to 20 of the first interview schedule for mothers (Appendix I) and on pages 3 to 5 of the interview schedule for fathers (Appendix III.) Where appropriate specific

questions will be quoted in the text or exact references will be given to the relevant question numbers. This practice will be followed throughout this section.

1. The Diagnosis of Cystic Fibrosis

The children varied considerably in age at the time of their diagnosis. The youngest to be diagnosed were the children born with bowel blockages which required surgical intervention in the first days of life. The oldest child to be diagnosed was 14 years old when cystic fibrosis was recognised. The mean age of the children in the sample at diagnosis was 2:1 years (S.D. of the age distribution when expressed in months = 35)

46 of these 58 children were diagnosed at the Royal Hospital for Sick Children in Edinburgh and 30 of them were diagnosed by Dr. McCrae.

From the history which mothers gave, of events which transpired between their first suspicion that the child was unwell and the diagnosis of cystic fibrosis, it was possible to code the degree of difficulty which they had encountered before obtaining this diagnosis (Q96 mothers' schedule) This coding scheme is best explained by examples:

Great difficulty: This is exemplified by a long period of referrals from doctor to doctor and characterised by conflicting diagnoses. It was considered very traumatic by the mother.

e.g. From the age of 6:0 years S's health began to deteriorate. He had constant diarrhoea and was rapidly losing weight. He had developed a bad cough. The family doctor diagnosed tuberculosis and referred the child to a

specialist clinic. There the specialist diagnosed bronchitis and referred the child to a 'Chest Unit' in a local hospital. The child was admitted, discharged and readmitted to this hospital without further diagnosis being made. The child was transferred to another hospital in the city when bronchitis was ruled out. A physician from the Royal Hospital for Sick Children was called in to consult on the case, and he diagnosed cystic fibrosis. The child was by then 7:0 years old and critically ill.

Some difficulty: This is then represented by a less traumatic history of shorter duration.

e.g. R. presented feeding difficulties from birth. These became particularly troublesome from the time when the child was taken home from hospital. The child vomited after each feed and, consequently, was not thriving. The family doctor, who was called several times, insisted that there was nothing wrong with the baby and refused to take any action. Another doctor in the same practice came on one occasion. He diagnosed bronchitis and prescribed some medicine. When it was ineffective, the family's doctor was called again and the child's grandmother then insisted that the child be referred to a specialist. R. was referred to the Royal Hospital for Sick Children and diagnosed at the age of 0:3 year.

Little or no difficulty: This is exemplified by the children who were diagnosed in the first days of life, either by presenting with meconium ileus

or by having a sibling already diagnosed. A few children, outwith these two categories were also diagnosed, with a minimum of difficulty.

e.g. J. (aged 2½ months) was not thriving and had frequent bouts of diarrhoea. The family doctor was called. He did not suggest a diagnosis but prescribed medicine (presumed by the mother to be treatment for the diarrhoea). The doctor returned frequently, without the mother having to ask, and, when a second prescription was as ineffective as the first, the child was referred to the Royal Hospital for Sick Children and diagnosed, by the age of 0:4 year.

Assessed in this way, the experience of at least some difficulty before obtaining a diagnosis of cystic fibrosis was found to be unexceptional. Table III.B(i).

Table III.B(i) The Incidence of Difficulty in Obtaining a Diagnosis of C.F.

	Mothers' report
Experienced great difficulty	40%
Experienced some difficulty	24%
Experienced little or no difficulty	36%

Total N refers to 58 diagnoses of C.F.

The time interval between the time of the diagnosis and the time of the enquiry varied from 8 months to 12:6years. The distribution of time intervals showed a mean of 4:11 years and a standard deviation of 3:0. The difficulty reported in obtaining diagnoses within the last four years was not significantly different from that reported in obtaining the diagnoses of children made more than four years ago ($X^2 = 2.08$, d.f. = 2, $P \simeq 0.30$). When one recalls that three of the children in the sample required urgent post-natal attention and that 9 of the children had the diagnostic advantage of a sibling already recognised as having cystic fibrosis (1 recognised at post-mortem), then the record looks serious indeed.

Although there were perfectly legitimate medical reasons for some of these delays, it was interesting that the mothers' reports of their experiences at this time suggested two variables which might also be significant in the degree of difficulty which was experienced in getting the diagnosis. These were the factors of the mother's age at the time of the diagnosis and the ordinal position of the child. Young mothers, who were under 23 years of age at the time of the child's diagnosis experienced significantly more difficulty in obtaining a diagnosis than did older mothers. ($X^2 = 19.2$, d.f. = 2, $P < 0.001$) However the difficulty in obtaining a diagnosis for the first born child in the family was not significantly greater than the difficulty experienced with later born children ($X^2 = 1.58$, d.f. = 2, not statistically significant.)

There were no social class differences in the reported degree of difficulty, but mothers whose children were diagnosed

at the Royal Hospital for Sick Children in Edinburgh tended to report less difficulty than mothers whose children were diagnosed elsewhere. ($X^2 = 5.58$, d.f. = 2, $P < 0.1$)

Only 4 of the mothers felt that they blamed themselves for the delay in the child's diagnosis whereas in 24 of the 58 cases mothers blamed the doctors, either the family doctor, hospital doctors or both, for not recognising the child's condition sooner. (Q97 and 98). This blame was fairly evenly distributed between doctors in these two spheres. Table III.B(ii)

Table III.B(ii) Mothers' Blame of Doctors for Delay in Child's Diagnosis

	Mothers
Blames family doctor	42%
Blames hospital doctors	37%
Blames both	21%
Total N = 24 diagnoses i.e. 41% of total no. of cases	

The permanent damage done by experiences at this time was recorded by asking mothers whether they felt their attitude to the medical profession had changed in any way. (Q102). Happily, only 18% of all the mothers reported a change for the worse, 14% on the contrary, reported an increase in their respect for the profession, while the remainder, 68%, reported no lasting change to their attitude in either direction, as a result of experiences at the time of the child's diagnosis.

Dr. McCrae had, from the first, expressed concern about the way in which a diagnosis of this nature should be managed, from a doctor's point of view and so parents' impressions of their experiences, an important aspect of his concern, were carefully documented. Firstly, it was important to ascertain to whom the diagnosis had first been given (Q95). Mothers' responses are tabulated below. (Table III.B(iii))

Table III.B(iii) To Whom was the Diagnosis of C.F. first given

To mother alone	48%
To father alone	12%
To both parents	34%
To mother + supportive other	2%
Other than to child's parents	2%
To no-one	2%

(Total N refers to 58 C.F. diagnoses)

Mothers were also asked about the access which they had had to the diagnosing physician after the diagnosis, and the use which they had made of such opportunities as had been available, for asking further questions. (Q95) This information is scored in Table III.B(iv)

Table III.B(iv) The Availability of the Diagnosing Physician, after the Diagnosis, and the Response of C.F. Parents.

Diagnosing physician not available/ not seen to be available	24%
Physician created facility for discussion which parents did not take up	23%
Physician available, parents went to him to ask further questions about the diagnosis	53%
N = 58 diagnoses	

It was necessary then to ascertain what parents understood of their children's illness, as explained to them at this time, and to assess their reactions to the manner in which the diagnosis had been communicated to them. The responses recorded here are derived from Questions 103-118 of the mothers' first interview schedules and from Questions 23-36 of the fathers' schedules, for 49 mothers and 46 fathers. Two of the three stepfathers were included in this assessment since they had been part of the family at the time of the child's diagnosis. The other stepfather had not, so, although he was interested in and informed about C.F. his responses are omitted from this section of the results. The other cases missing from Table III.B(v) are those parents who were not available for interview.

Table III.B(v) Information given to Parents at the Time of the Diagnosis of Cystic Fibrosis - Parents' Reports.

	Mothers	Fathers
Was told that C.F. is inherited	88%	76%
Was told that it is chronic and incurable	100%	89%
Was told it might get worse in future	57%	50%
	N = 49	N = 46

Parents were asked what they had been told at the time when their children were first diagnosed as having cystic fibrosis. (Table III.B.(v)) They were also asked whether trouble had been taken to explain the diagnosis to them. The percentage of parents who reported that no-one had taken trouble to explain the diagnosis to them thoroughly, and the percentages of parents who, on the contrary, commend their family doctor or the hospital consultant in this respect, are shown in Table III.B(vi). (Some parents mentioned both doctors in their praise so the figures quoted sum to more than 100%)

Table III.B(vi) Who Took Trouble over the Explanation of the Diagnosis to Parents?

	Mothers	Fathers
No-one	45%	59%
The hospital consultant	49%	39%
The family doctor	8%	6%
	N = 49	N = 46

Overall 43% of mothers and 35% of fathers expressed complete satisfaction with the way in which the diagnosis had been communicated to them, the remainder were more critical of the way in which the situation had been managed. The most common complaint was that the prognosis had been painted too black and that the news had been broken too bluntly, leaving them no room for hope. A few parents made comments in direct opposition to this i.e. that the diagnosing physician had tried to break the news to them too gently so that they had, for some considerable time, failed to appreciate the seriousness of the situation. This was not a criticism which was ever levelled at clinicians in the Royal Hospital for Sick Children.

Parents were asked whether they thought the doctor should tell parents what he suspected was the matter with their child or whether he should delay telling them until his diagnosis had been confirmed by objective tests. Interestingly the mothers were almost equally divided on this question (Q113) with the casting vote falling on the side of being told what the doctor suspected.

Parents' responses to the communication were naturally those of shock, although parents who had experienced a great deal of difficulty in obtaining the diagnosis did say that, in a way, they also felt relief that the child at least had something that the doctors knew how to treat. Feelings of disbelief were only momentary as were the feelings of resentment but the duration of the feeling of having been stunned was of a variable duration which was hard to assess.

Mothers, who were clearly made highly anxious by the diagnosis of cystic fibrosis, were asked about their main worry at this time (Q95) 78% of them reported, unhesitatingly, that their dominant fear then was that their child was about to die. 16% of mothers were primarily anxious about their own ability to cope, while the remainder expressed worries about the quality of life in store for their child with cystic fibrosis.

Information-seeking behaviour has often been described in the literature as a characteristic phase of parents' coping response to such a diagnosis for their child e.g. by Hamburg and Adams (1967.) It was interesting then, to observe that as many as 22% of these mothers and 32% of fathers had not actively sought information since their child's diagnosis. Rather a large proportion of parents, 64% of mothers and 59% of fathers, was, however, still desirous of more information about cystic fibrosis at the time of the enquiry.

Some questions had been included in the schedule to assess the means by which parents had obtained or would have liked to have obtained such information.

Unfortunately, the question which was concerned with the

role of the C.F. Research Trust in providing this information, was the subject of some confusion. The C.F. Trust circulates two kinds of publication for parents. The first takes the form of a series of pamphlets written by specialists to explain selected aspects of cystic fibrosis or its treatment to lay readers e.g. "Physiotherapy in Cystic Fibrosis, Why-How-When". It was to those informative pamphlets that our questions referred (Q115 of mothers' first questionnaires and Q33 of fathers' schedules). However these pamphlets had not been widely distributed among the families in this sample and confusion arose between these and the more widely distributed Newsletter. This latter monthly publication of the C.F. Trust, reports primarily on fund-raising activities and of the exploits of young people who have the disease. Although it does carry reports of ongoing research, it is fair to say its informative function in this respect is secondary to its morale-boosting aims.

This confusion was detected rather too late in the study for it to be effectively remedied, although this did at least offer the consolation that even had a pilot study been possible such confusion might still have passed undetected. For this reason, though, the estimated 69% of mothers who reported having seen the C.F. Trust pamphlets and having been helped by them, is probably too high.

There was no confusion however, about the suggestion that the diagnosing physician might provide some written information for parents to take home with them, after he had explained the diagnosis to them personally and 84% of mothers and 67% of fathers were enthusiastic about the possibility.

Parents were asked how much a number of different sources had helped them in understanding cystic fibrosis. (Mothers' first schedule, Q118; Fathers' schedule, Q36.) Their responses were coded as follows:

Not at all	=	0
Very little	=	1
Moderately	=	2
Very much	=	3

A total score was obtained for each possible agent of help and, in order to provide uniformity of assessment between mothers' and fathers' scores, the mean score assigned to each was calculated. The agents of help attaining mean scores greater than 0.5 are listed in Table III.B(vii). The mean score assigned to them by 49 mothers and, now, 47 fathers, is given in parentheses.

Table III.B(vii) Parental Aids in the Understanding of Cystic Fibrosis (in order of decreasing helpfulness)

Mothers (mean score)		Fathers (mean score)	
1. Clinic doctors	(2.5)	1. Clinic doctors	(2.2)
2. Family doctor	(1.3)	2. Books	(1.1)
3. Physiotherapist	(1.0)	3. Family Doctor	(0.8)
4. Books	(0.9)	4.(C.F. Parents Group	(0.6)
5.(C.F. Parents Group	(0.6)	(Other parents	(0.6)
(Other parents	(0.6)		

Other suggested agents of help to parents in this matter, friends, family, clergymen and social workers, were found helpful by only a small minority of parents.

Bearing in mind the background against which these diagnoses of cystic fibrosis were made, it was felt that an index could be derived, from the information already available, of the degree of parents' satisfaction with the clinical management of the diagnosis of their child. The derivation of this index, which may be important in understanding parents' later attitudes and behaviour, is explained in Table III.B(viii).

Table III.B(viii) To Derive an Index of Parental Satisfaction with the Management of their Child's Diagnosis

If the conditions, as stated, are satisfied the number of points as indicated would be added to the total value of the Index.	
1. The diagnosis should have been given to both parents together. (Unless in exceptional circumstances)	1
2. There should have been more than one opportunity for parents to discuss the diagnosis with a doctor	1
3. Care should have been taken in the diagnosis (1 point for each of mother's and father's views)	2
4. Parents should have been satisfied that the diagnosis was communicated to them as well as possible in the circumstances (1 point each)	1
5. Parents should have had maximal help in understanding C.F. both from the family doctor and from the hospital physician. (0-3 points for help given to each parent by each doctor)	12
Range of Index Values = 00-18 points	

On the basis of parents' reports then, the values of this index varied from 1 to 18 and showed a mean value of 9.3 with a standard deviation of 3.6. There was no evidence of a statistically significant difference between the indexed levels of satisfaction of parents whose children were diagnosed more that four years ago and those of parents whose children were diagnosed more recently ($X^2 = 1.59$, d.f. = 2, $P \doteq 0.50$.)

Since observations have been made in the literature (e.g. by Blumenthal, 1969) that the management of the communication of the diagnosis of cystic fibrosis to parents, is an important factor in the volution of parents' subsequent attitudes and behaviour, this index may prove useful in the interpretation of later findings.

2. Parental Comprehension and the Genetic Implications of Cystic Fibrosis.

In considering the management of the diagnosis from the parents' viewpoint we had already scored in a simple binary way, the information which parents thought they had, or had not, been given by the diagnosing physician. However it was necessary to assess further how well these parents understood the disease at the time of the enquiry, in order to understand their ongoing attitudes and behaviour recorded at that time.

Parental comprehension of the nature of cystic fibrosis was coded from the responses of parents to several of the questions discussed in the preceding part of the chapter (Mothers' first schedule Q103 - 107 and Q111) (Fathers' schedule Q23 - 26, Q29.) From their responses to these questions parents had indicated the

extent of their awareness of the nature of the disease as well as of its being inherited, chronic, incurable and potentially fatal and their comprehension was then readily graded good, moderate or poor. The responses of 49 mothers and 47 fathers were graded in this way to give the distribution of scores shown in Table III.B(ix).

Table III.B(ix) Parental Comprehension of the Nature of Cystic Fibrosis

	Mothers	Fathers
Showed good understanding of C.F.	38%	32%
Showed moderate understanding	44%	21%
Showed little or no understanding	18%	47%
	N = 49	N = 47

It was suspected that the level of parental comprehension might reflect the parents' intelligence and the number of years of formal education which they had had. In order to estimate the strength of any such relationship the graded assessments of parents' understanding were correlated with their score on Cattell's factor B, and with their school leaving age. The values obtained for Pearson's r, and the significance levels of these values, for these correlations are shown in Table III.B(x). These significance values represent the chances that their respective

correlation coefficients for the sample, could deviate by as much as they do, from an hypothesised population correlation of zero (Guilford, 1965.)

Table III.B(x) Values of r, and their significance, for the Correlation of Parental Understanding of Cystic Fibrosis with Parental Intelligence and Education

	Mothers		Fathers	
	r	Significance level	r	Significance level
School leaving age	0.46	0.001	0.46	0.001
Intelligence	0.23	0.05	0.16	0.1
	N = 49		N = 47	

It was suspected that the level of understanding of the disease shown by parents at the time of the enquiry might reflect the way in which the diagnosis had been managed. To test this hypothesis, correlations were obtained of the index of parental satisfaction with the management of the diagnosis, as calculated, with parents' comprehension of cystic fibrosis. This correlation only reached statistical significance in the case of fathers.

(Mothers - r = 0.18, significant only at the 10% level;
Fathers - 4 = 0.35, significant at the 0.1% level)

One important aspect of this assessed level of parental understanding requires separate attention however, and this

concerns parents' comprehension of the genetic nature of cystic fibrosis. It has already been recorded (Table III.B(v)). That the majority of parents had some degree of comprehension of the fact that cystic fibrosis is an inherited disease but a substantial proportion of parents felt that its genetic basis had never been fully explained to them. Table III.B(xi)

Table III.B(xi) Parents Report of Information given about Genetics of C.F. at Diagnosis

	Mothers	Fathers
Not told that C.F. is inherited	12%	23%
Genetics not fully explained	39%	43%
	from N = 49 mothers	from N = 44 fathers

Since this aspect of the information concerns only the natural parents of the C.F. child, i.e. those parents who are themselves C.F. carriers, the reports of stepfathers are not included in this section of the results.

The parents' actual understanding of the genetic nature of the disease was gauged from their response to the question "What do you understand of the chances of your children's children having cystic fibrosis?" (Q109 on the mothers' first schedule and Q27 on the fathers' schedule.) Understanding was then rated

as 'good' when the parents appreciated fully the nature of recessive inheritance, that their affected sons were probably sterile that their affected daughters had an increased chance of bearing an affected child and that their phenotypically well children could be carriers of cystic fibrosis. Moderate understanding was credited when some, but not all of this information had been grasped. Understanding was rated as poor when the parents knew simply that their child's illness had been inherited and that there was some chance that any other child conceived in the future might also be affected. There was a high degree of correspondence between the distributions of grades assigned to mothers and fathers using this scheme (X^2 = 41.7, d.f. = 9, $P < 0.001$) so the distribution quoted shows the extent of understanding over the sample as a whole. This is compared with the understanding shown, of the general nature of cystic fibrosis, in Table III.B(xii).

Table III.B(xii) To compare the levels of parental comprehension of C.F. in general, with those of the genetic aspects in particular.

	of C.F. in general	of the genetic aspects of C.F.
Show good understanding	35%	22%
Show moderate understanding	33%	23%
Show little or no understanding	32%	55%
	N = 96	N = 93

Again, the level of understanding of the genetic aspects of C.F. shown by parents showed a fairly high level of correlation with the parents' school leaving age (Mothers, $r = 0.42$, $P < 0.001$; Fathers, $r = 0.57$, $P < 0.001$) and a slightly less significant degree of relationship with parents' scores on Cattell's Factor B (Mothers, $r = 0.26$, $P < 0.02$; Fathers, $r = 0.19$, $P < 0.07$). Thus parents who were less intelligent and those who had had fewer years of formal education were less likely to achieve a high level of understanding of the inheritance of cystic fibrosis.

Mothers who showed a high level of understanding of the nature of the disease did not seem any more likely than the others to show a high degree of comprehension of its genetic aspects ($r = 0.05$, $P \simeq 0.35$) whereas a strong positive association was found between the levels of fathers' understanding of these two aspects of cystic fibrosis ($r = 0.31$, $P < 0.01$). Fathers' level of understanding of the genetic aspects also bore a significant relationship to the way in which the diagnoses of their children had been managed. A high degree of satisfaction with the diagnosis management indicated that the father had been included in the initial explanations of the disease, and this inclusion of the father in the diagnosis was found to be positively associated with his level of comprehension of the genetic basis of the disease ($r = 0.33$, $P < 0.05$).

In view of these genetic implications of cystic fibrosis, family planning advice might have been offered, but it was found that only about one third of the parents in the sample had ever had this aspect of the problem discussed with them by a doctor (Table III.B(xiii)). It is interesting to note that this seemed

to be a social-class related phenomenon. Family planning had been omitted from discussions with 64% of the families representing social classes I and II, whereas it had been omitted from discussion with only 28% of social classes III to V.

Table III.B.(xiii) The Absence of Discussion on Family Planning

	Mothers	Fathers
No discussion with a doctor	36%	32%
No discussion with anyone	18%	16%
	N = 49	N = 44

Families whose children had been diagnosed in the Royal Hospital for Sick Children were compared with those whose children had been diagnosed elsewhere. No significant difference was found between the groups, in the proportions of parents who had discussed family planning with their doctors (X^2 = 0.001, d.f. = 1) nor was the matter any more or less likely to be discussed if the family had attended Dr. McCrae rather than any other doctor. There were only 4 Roman Catholic couples in the sample, providing a very restricted basis for comment about the extent of religious differences in the incidence of medical advice offered, but doctors had discussed the question of family planning with parents in each of these families.

In approximately half of the cases where there had been no discussion of family planning with a doctor, it was found that there had been no discussion of this subject with anyone, not even between spouses (Table III.B.(xiii)) and again this was a social class-related effect. Discussion on the subject occurred between parents in all but one of the families in S.E.S. groupings I and II (the exception being the case of a C.F. child born as a late baby; a further pregnancy was assumed to be impossible.) In the homes of families of lower social status, if family planning was not discussed by the doctor, it was unlikely to be discussed at all.

Although the findings relating to the more subtle implications of the genetics of cystic fibrosis are more appropriately recorded in a later chapter, (Section III.D) the results referring to the more immediate behavioural aspects of parents' response to this information are included in this chapter. The whole issue of genetic counselling in cystic fibrosis will be drawn together in the discussion (Section IV.)

Following the first diagnosis of cystic fibrosis in the family the majority of couples decided that they should have no further children. However, their responses to the question "Do you think that having N (name of child) has changed your feelings about having more children in any way?" revealed that the consideration of the family size desired by the couple was as important in this decision as the diagnosis of cystic fibrosis (Table III.B.(xiv))

Table III.B.(xiv) Family Planning After the Diagnosis of Cystic Fibrosis

Family to be limited because of cystic fibrosis	36%
Family to be limited because of family size	36%
Further pregnancies intended	28%
N = 50 families	

On the whole then, the influence of cystic fibrosis on family planning was not as great as might have been anticipated. The chances that parents will continue to act, post-diagnostically, as they planned, prediagnostically, are high; the chi-square value for the association between parents' intentions and subsequent behaviour is significant at the 1% level ($X^2 = 7.24$, d.f. = 1). The relationship between parents' understanding of the genetic risk and their family planning decision does not reach statistical significance, (Mothers $X^2 = 2.73$, d.f. = 3, $P \simeq 0.5$; fathers $X^2 = 5.79$, d.f. = 3, $P \simeq 0.1$) nor does the parents' realisation of the fact that the disease is potentially fatal seem to exert significant influence over their decisions in this matter (Mothers $X^2 = 3.13$, d.f. = 1, $P \simeq 0.1$; fathers $X^2 = 1.38$, d.f. = 1, $P \simeq 0.3$).

The stability of this decision, at least from the mothers' viewpoint, is to some extent endorsed by the finding that, given the hypothetical opportunities of obtaining intra-uterine diagnosis and selective abortion, the number of mothers who said they would

then be willing to risk further pregnancies in the hope of having a well child was fairly small, representing 22% of the families in the sample.

The outcome of these decisions clearly raised the question of birth control. Parents were simply asked whether this was an area in which they required help (Q123 and 124 of mothers' first schedule). For two of the 4 Roman Catholic families in the sample this issue constituted a serious problem on religious grounds. The remaining women who requested help of this kind were not Roman Catholics but were three who wished to be sterilised. The numbers of mothers who actually asked for help with contraception is clearly an underestimate of the number who need help, since the number of 'accidental' conceptions was rather high. The outcome following parents' family planning decision is indicated in Table III.B.(xv).

Table III.B.(xv) Outcome of Family Planning Decision.

(a) Parents intended to have no further children	(N = 36 families)
Sterilisation	- 10
Vasectomy	- 2
Accidental Pregnancy ending in - Miscarriage	- 3
- Termination	- 4
- At term	- 2
(b) Parents intending to have further children	(N = 14 families)
No. of further children born	- 21
No. of these having C.F.	- 3

Before concluding this report of the findings immediately relevant to the genetic aspects of cystic fibrosis, it is interesting to speculate about the influence of medical discussion on the decisions made by parents in this context. The results show that, of those who had discussed family planning in the light of cystic fibrosis, with a doctor, almost 33% had no further children in spite of their previous prediagnostic intentions to do so. Almost all of the remaining 67% continued to have, or to refrain from having, further children in accordance with their original intentions. (The word 'almost' is inserted to account for accidental pregnancies.) Among parents who had had no such discussions with a doctor only 1 in 6 altered their original family planning in the face of the diagnosis, the remaining 5/6ths carried out their original intentions.

3. Medical Supervision

Continuing in its concern to describe the medical management of cystic fibrosis, the study then sought to gather information about parents' attitudes to the system of care provided for their children. Since it was observed that mothers were primarily responsible for bringing children to a doctor's surgery or to an out-patient clinic, the findings reported here reflect questions which appeared only on the interview schedules for mothers (Q125 - 150). Attitudes to family doctors were investigated first.

Mothers were asked to whom they would refer if they required medical attention for, or advice about, their child's condition and 60% of mothers said they would go first to their G.P. as against 38% who said they would rather consult the hospital specialist

directly. There is, however, reason to believe that this pattern underwent a change during the course of the enquiry at least for patients in the Edinburgh area, so that these figures cannot be interpreted too literally.

In spite of mothers' expectation that they would contact the family doctor in the case of difficulty with the C.F. child, the reported frequency of contact with the G.P. for that child was lower than might have been expected. Table III.B.(xvi).

Table III.B.(xvi) Frequency of Contact with Doctors re: Cystic Fibrosis

	G.P.	C.F. Clinic
Once per month - once per 2 months	30%	76%
Once per 3 months - once per 6 months	58%	10%
Less than once every 6 months	-	8%
Very rarely/never	12%	6%
	N = 50	N = 50

Frequency of contact with the general practitioner reflected most clearly the child's age. Children under the age of five years were most likely to require his attention most frequently, although, at this stage, it was difficult for mothers to distinguish which problems were in fact, due to the child's cystic fibrosis.

From the information gathered it was possible to derive an index of mothers' attitudes to their family doctors. Table III.B.(xvii)

Table III.B.(xvii) Index of Mothers' Attitudes to G.P.

1. The C.P. was not held responsible, by the mother, for any delay in the diagnosis of C.F. in her child (Q.98)	1
2. The G.P. had been instrumental in the mother's understanding of C.F. (Helpfulness scale, scored by mother 0-3; Q.118)	3
3. The G.P. was reported to be generally helpful to the mother in the care of the C.F. child (Q.127)	1
4. Mother expressed confidence in the G.P.'s understanding of cystic fibrosis (Q.127.b.)	1
Total Score	0-6

This index could then take values from 0, representing a very negative attitude on the part of the mother, to the G.P., to 6. The value 6 represented a good working relationship between the mother and the family doctor. The distribution of index values thus obtained is tabulated below. Table III.B.(xviii).

Table III.B.(xviii) Distribution of Indices of Attitudes to G.P.'s.

Index Value	
0 - 1	24%
2 - 4	42%
5 - 6	34%
Mean value = 3.4	N = 50

Families of lower social classes seemed to hold much more favourable attitudes to their family doctors in this respect, than did those of higher socio-economic status ($X^2 = 10.65$, d.f. = 2, $P < 0.01$). Not surprisingly those attitudes bore heavily on parents' behaviour, and parents who had a good relationship with their family doctor were more likely to consult him first when they were anxious about their C.F. child ($X^2 = 18.81$, d.f. = 2, $P < 0.001$).

A common problem when a patient attends more than one doctor, and when the doctors are geographically and administratively separate, can be that of communication between the doctors about the patient. C.F. families were asked about the effectiveness of this communication, in their experience, and about any cases of confusion or lack of agreement which had occurred. Long delays were often reported between the recommendations for treatment made at the clinic and the acknowledgment of these recommendations by the G.P. and confusions and disputes were not unknown. On the

basis of parents' answers to questions 144 and 145 about delays and difficulties, it was possible to score the communication between their G.P. and the hospital on a three point Scale:

No difficulty = 0

Some difficulty = 1

Considerable difficulty = 2

36% of these mothers reported having experienced no difficulties of this kind, while 24% had had some difficulty and 40% had had considerable difficulty caused by delays and confusions along this channel of communication. These communication difficulties presented a number of practical problems to mothers as well as, in some cases, undermining their confidence in the family doctor.

Turning, then, to the system of out-patient care, we find that differences exist between the patterns of hospital-based care provided for the Royal Hospital for Sick Children, (R.H.S.C.) and elsewhere. Among the eight families in the sample whose children were not patients at R.H.S.C., the condition of the C.F. child was reviewed at least every two months, in only one case. The remaining seven cases attended hospital clinics less frequently. Among the remaining 42 families attending R.H.S.C., 37 had an appointment at the C.F. clinic at least every eight weeks, although most of these children were seen at monthly intervals. This difference may be important in the interpretation of the results.

Such regular medical supervision of the children from a hospital out-patient clinic, required investigation from the point of view of practical convenience for the families, as well as in terms of medical management. A rough estimate was made of the distance between the families' homes and the clinic which they attended (Table III.B.(xix).

Table III.B.(xix) Distance of C.F. Homes from C.F. Clinic

Within walking distance	- 8%	15 miles - 28%	
Across town/city	- 26%	30 miles - 28%	
		> 30 miles - 10%	

N = 50 families

The corollary to this question was then to ask how the families negotiated this distance for clinic visits. The pattern of usage of different modes of transport for this purpose is indicated in the next table (Table III.B.(xx), from Q.78.)

Table III.B.(xx) Modes of Transport to C.F. Clinic

On foot	- 6%	W.V.S. car	- 2%
Private car	- 34%	Ambulance	- 22%
Public transport (bus or train)	- 32%	Taxi	- 4%

N = 50 families

It was encouraging, however, to find that only one family reported that clinic attendance created a continuing source of financial difficulty. A further eight families reported that financial hardship in this connection, occurred only sometimes. Clearly these families sought help, either financial or practical with this problem. Other families who said they would

like help with the transportation of their C.F. children to the clinic, a further five families, sought practical rather than financial assistance, e.g. because the child was very young or in poor health. Aid of this sort was felt to be most needed in bad weather.

Other important factors in parents' experience of attending clinics concerned the time factor of which three relevant facets were investigated. Parents were asked to estimate how long they were away from home for clinic visits, how long they had to wait before seeing the doctor and how long they spent with him (Q.131 - 133). Their responses are subsumed under Table III.B.(xxi).

Although 26% of the mothers felt that they would like to spend longer with the doctor at these visits (Q.134) their satisfaction, or lack of it, bore little relationship to the estimates the mothers had given of the time that they already spent in the consulting room ($X^2 = 3.55$, d.f. = 4, $P \simeq 0.50$).

Table III.B.(xxi) The Time Factor in Attending C.F. Clinics

(a) Parents' estimates of time spent away from home for Clinic Visits

⩽ 1 hr.	⩽ 2 hrs.	⩽ 3 hrs.	⩽ 4 hrs.	> 4 hrs.
-	30%	32%	14%	24%

(b) Parents' estimates of time spent waiting to see the doctor

≃ 15 mins.	≃ 30 mins.	≃ 45 mins.	≃ 60 mins.	> 60 mins.
60%	26%	8%	6%	-

(c) Parents' estimates of time spent with the doctor

⩽ 5 mins.	⩽ 10 mins.	⩽ 15 mins.	⩽ 20 mins.	> 20 mins.
8%	28%	46%	14%	4%

N = 50 families

The third and final matter of clinic administration which was very relevant to these families was the more personal one of continuity of care, i.e. whether the family saw the same doctor at each clinic visit and indeed whether this was important to them. Only 16% of the 50 families said they saw the same doctor at the clinic each time they went, whereas 68% of the remainder said that they found it confusing to have no regular appointment with the same doctor for each visit, and would have liked to have seen the same person each time. Reasonably enough, not all expect this person to be the consultant physician (although many do), but several parents expressed the desire to be able to make an appointment specifically with him, if they were anxious or the

child was ill. These parents were not aware that it was possible to do this under the existing system of clinic administration.

These practical aspects of the accessibility, time commitment and the appointment system of the clinic were thought to be rather closely related to parents' regularity of attendance at the clinic and to their attitudes to this aspect of the medical supervision of their C.F. children.

At the end of the study, the regularity with which the children had been brought to the clinic, relative to the number of appointments they should have had was assessed from their case notes on a 4-point scale:

never/lapsed	- 0	fairly regular	- 2
rare attendance	- 1	very regular	- 3

In this way the differences in practice between the R.H.S.C. and other hospitals were ironed out.

The regularity with which appointments at the clinic were kept was related to a significant degree to the distance of the family's home from the clinic, and to whether clinic attendance created financial difficulties for them. Curiously though, the difference in reliability of families who had to pay for their transportation was not significantly different from that of those who did not. The time factor and the lack of personal continuity of care at the clinic did not significantly affect the attendance record of the families. Chi-square values are given in Table III.B.(xxii) with the pattern of attendance of these families at the clinic.

Table III.B.(xxii) Factors in the Regularity of Families' Attendance at the C.F. Clinic

Regularity of Attendance:	0 never/lapsed	1 rare	2 f.reg.	3 v.reg.
% of 50 families :	3%	17%	16%	64%

Factors in their Attendance:	X^2	d.f.	P
Distance from Clinic	6.05	2	< 0.05
Mode of transport	3.70	2	$\simeq 0.20$
Financial hardship	13.6	6	< 0.05
Time commitment (away from home)	5.70	3	$\simeq 0.10$
Satisfaction with time spent	2.70	3	$\simeq 0.50$
Maternal comprehension of C.F.	18.81	1	< 0.01

Mothers who regularly brought their children to the clinic were found to be siginificantly more likely to obtain higher scores for their comprehension of cystic fibrosis. This finding leads to the rather circular argument of whether the better informed mother is more likely to attend the clinic more regularly or whether more information is one of the rewards which accrues from regular clinic attendance. As in many cases of this kind, both suggestions probably hold some truth.

From the remaining responses which parents had given to questions relating to the medical supervision of their children from

the clinic, it was possible to tease out those which reflected their attitude to the doctors whom they saw there (Q135 - 137, Q142 - 143), and those which were, rather, indicative of their attitude to the way in which the clinic was run (Q138 - 141, Q148). If the value 1 is assigned to 'Yes' responses, except in Questions 142 and 148 where 'No' responses score 1, then a five-point scale can be derived for each of these attitudes. Scores on this scale were interpreted as follows:

Negative, highly critical attitude, scores = 0 - 1

Some satisfaction, some criticism, scores = 2 - 3

Positive, highly satisfied attitude, scores = 4 - 5

Table III.B.(xxiii).

Table III.B.(xxiii) Indices of Attitudes to the C.F. Clinic

a. Relationship with clinic doctors

Q.135. Do you feel you can tell him (the doctor) all your worries?

Q.136. Do you feel he really understands the child?

Q.137. Do you feel he can really help you with treatment?

Q.142. Have you ever felt angry towards the doctors in the clinic?

Q.143. Would you say the clinic doctor has been a lot of help to you?

Total Score for Responses, 0 - 5

b. Attitude to the way in which the Clinic is run.

Q. 138. Can you always get in touch with the doctor when you want to?

Q. 139. Do you like the way the clinic is run?

Q. 140. Do you like meeting mothers of other C.F. children in the clinic?

Q. 141. Do you think N likes meeting the other C.F. children?

Q. 148. Would you like to change the clinic arrangements in any way to improve them?

Total Score for Responses, 0 - 5

On this basis there was a high degree of correspondence between the parents' attitudes on the two scales, suggesting some degree of spread of these attitudes. If parents were satisfied with their relationship with the clinic doctors they were more likely to express satisfaction with the way in which the clinic was run and vice versa ($X^2 = 10.2$, d.f. = 4, $P < 0.05$). The distributions of scores and their mean values are indicated in Table III.B.(xxiv.)

Table III.B.(xxiv) Indices of Parental Attitudes to the C.F. Clinic

	Re: clinic doctors	Re: clinic administration
Critical Attitude	8%	8%
Mixed reaction	32%	42%
Highly satisfied Attitude	60%	50%
	N = 50 families	

the clinic, it was possible to tease out those which reflected their attitude to the doctors whom they saw there (Q135 - 137, Q142 - 143), and those which were, rather, indicative of their attitude to the way in which the clinic was run (Q138 - 141, Q148.) If the value 1 is assigned to 'Yes' responses, except in Questions 142 and 148 where 'No' responses score 1, then a five-point scale can be derived for each of these attitudes. Scores on this scale were interpreted as follows:

Negative, highly critical attitude, scores = 0 - 1

Some satisfaction, some criticism, scores = 2 - 3

Positive, highly satisfied attitude, scores = 4 - 5

Table III.B.(xxiii).

Table III.B.(xxiii) Indices of Attitudes to the C.F. Clinic

a. Relationship with clinic doctors

Q.135. Do you feel you can tell him (the doctor) all your worries?

Q.136. Do you feel he really understands the child?

Q.137. Do you feel he can really help you with treatment?

Q.142. Have you ever felt angry towards the doctors in the clinic?

Q.143. Would you say the clinic doctor has been a lot of help to you?

Total Score for Responses, 0 - 5

b. Attitude to the way in which the Clinic is run

Q.138. Can you always get in touch with the doctor when you want to?

Q.139. Do you like the way the clinic is run?

Q.140. Do you like meeting mothers of other C.F. children in the clinic?

Q.141. Do you think N likes meeting the other C.F. children?

Q.148. Would you like to change the clinic arrangements in any way to improve them?

Total Score for Responses, 0 - 5

On this basis there was a high degree of correspondence between the parents' attitudes on the two scales, suggesting some degree of spread of these attitudes. If parents were satisfied with their relationship with the clinic doctors they were more likely to express satisfaction with the way in which the clinic was run and vice versa ($X^2 = 10.2$, d.f. = 4, $P < 0.05$). The distributions of scores and their mean values are indicated in Table III.B.(xxiv.)

Table III.B.(xxiv) Indices of Parental Attitudes to the C.F. Clinic

	Re: clinic doctors	Re: clinic administration
Critical Attitude	8%	8%
Mixed Reaction	32%	42%
Highly satisfied Attitude	60%	50%
	N = 50 families	

Once more there were indications of a relationship, though not one reaching statistical significance, between attitude and comprehension of C.F. mothers who expressed satisfaction with their relationship with the clinic doctor(s) tended to be also those who were assessed as having a good understanding of cystic fibrosis ($X^2 = 3.26$, d.f. = 1, $P < 0.10$) Although there was a trend for parents of higher social class to express more favourable attitudes to the clinic doctors this did not reach statistical significance ($X^2 = 2.53$, d.f. = 1, $P \simeq 0.20$).

It was interesting that the computer analysis of the personality data suggested two further factors which were related to parents' attitudes as expressed here. There was evidence of a relationship between high scores on Cattell's Factor Q3 and positive attitudes to clinic doctors and between low socres on Cattell Factor Q4 and such attitudes. Thus mothers who have strong control over their emotions and general behaviour (high Q3) and those who are relaxed and composed (low Q4) seem more likely to be those who are able to report having achieved a satisfactory relationship with their child's clinic doctor (Q_3 : $X^2 = 10.67$, d.f. = 4, $P < 0.05$; Q_4 : $X^2 = 10.02$, d.f. = 4, $P < 0.05$).

4 Treatment

The conscientiousness with which the prescribed treatment regimen is followed in the child's home is of paramount importance if his disease is to remain under satisfactory medical control. For this reason, considerable attention was given to the

administration of this fairly strenuous programme of therapy in the home and parents' responses to Q.151 - 199 of the first interview schedule are summarised here. The findings for 58 children are quoted, since the treatment routine specified, varies from child to child, even two children in one family are likely to differ in their therapeutic needs.

Parents were asked to specify the kinds of treatment which their children required. Table III.B.(xxv) indicates just how strenuous the therapeutic regime is likely to be.

Table III.B.(xxv) Number of Different Kinds of Treatment Prescribed for C.F. Children

1	Kind of treatment	7%
2-3	Kinds of treatment	59%
4	Kinds of treatment	34%
		N = 58 Children

The implications of these separate treatments were discussed in Section I in the 'layman's introduction to cystic fibrosis'. A simple reminder of the nature of the treatments to which the interview referred may, however, be obtained by reference to Table III.B.(xxvi) which lists the treatments mentioned in the order of decreasing nuisance value. This rank order was derived from the total number of mothers of these 58 children who had reported the treatment as being troublesome.

THE UNIVERSITY *of* EDINBURGH

PAGE MISSING IN ORIGINAL

Table III.B.(xxvi) Treatments for Cystic Fibrosis in order of Decreasing Nuisance Value

1. Physiotherapy	- 32%	4. More than one kind of treatment is troublesome	- 14%
2. Pancrex	- 24%		
3. No treatment is troublesome	- 17%	5. Antibiotics	- 7%
		6. Others	- 6%

refers to N = 58 children

Expressed in this way, of course, the frequency with which a particular kind of treatment is used, must influence the proportion of mothers who can then report it as being the most troublesome. There is little doubt that, in the past, the mist tent would probably have ranked equal in nuisance value to physiotherapy.

The nuisance value of Pancrex reported by these mothers is, on the whole, an objective comment on the real difficulties of supply and distribution to which this medicine has been subject. All that this table really shows, then, is that a high proportion of families (32% + 14%) find physiotherapy very troublesome.

The level of the child's protest was anticipated as being instrumental in determining the nuisance value of any treatment to parents. A global assessment of the degree of each child's protest was made from the information available about his response to the separate aspects of his treatment. A four-point scale of protest was applied:

Erratum - Due to an error in the numbering of the pages there is no page 206.

no protest - 0 moderate protest - 2

slight protest - 1 severe protest - 3

and scores were assigned on the basis of the number, duration and severity of the child's protests against therapy. Table III.B.(xxvii).

Table III.B.(xxvii) C.F. Children's Protests against Treatment

No protest	17%
Slight protest	34%
Moderate protest	27%
Severe protest	21%
N = 58 children	

Mothers had been asked about the age at which their children protested about each kind of treatment and from these responses it was possible to identify the age at which the child had been most difficult, with regard to his reaction to treatment, to date. The number of mothers quoting each of a number of age ranges, as being particularly trying, is quoted. Table III.B.(xxviii). This information is really only meaningful when compared with the proportions of children in the sample as a whole, who have reached or passed the age range in question.

Table III.B.(xxviii) To Identify whether C.F. Children Show Age Differences in their Degree of Protest against Treatment

	No. of Mothers who report this age most troublesome	Total No. of Children of this age group or older	∴ %age who find this age most trouble-some
< 3 yrs	5	58	5/58 = 9%
3 - 5 yrs	9	49	9/49 = 18%
5 - 8 yrs	11	38	11/38 = 29%
8 - 11 yrs	2	21	2/21 = 10%
> 11 yrs	5	8	5/8 = 62%
		Total N = 58 children	

Child is not troublesome over treatment: 10

Child has always been troublesome over treatment: 16

The amount of information which parents had given their children about each of the forms of treatment they required was scored on a very simple basis. If the child had been given at least some simple veridical explanation of why each treatment was necessary this was deemed adequate. If only some treatment had been thus explained the child's information was said to be incomplete. False information was easily identified, as was the category of 'no information given', although the children who were told only to do as they were told, or 'as the doctor says', were also included in this category. Although there are some of the youngest children for whom clearly this question is

irrelevant the scores for all 58 children are given in Table III.B.(xxix).

Table III.B.(xxix) Information given to Children about Treatment

No information given	34%
False/misleading information given	6%
Incomplete information given	26%
Adequate information given	34%
N = 58 children	

Caring for a cystic fibrosis child is a demanding task and, in many aspects of that task, the load falls heavily upon mothers' shoulders. Responsibility for supervising the child's daily treatment at home is often shared by fathers though, and the fathers of 25 of the 58 children were reported by the children's mothers to be willing to undertake responsibility for any or all of the child's treatment. Some fathers had never learned how to give physiotherapy so that the fathers of 17 of the 58 children were only willing to take responsibility for the other aspects of the child's treatment. The fathers of the remaining 16 children took no part in their treatment. However, the fathers of 6 of these children were not living in the same house as their families.

In order to assess just how demanding the task really is, mothers were asked to estimate the time which they, and their husbands, spent in the treatment of their C.F. child, in an average day. Table III.B.(xxx).

Table III.B.(xxx) Time Spent by Parents in C.F. Treatment, in minutes per day.

	Estimated Time Spent per Child, per day in mins.					
	0	≤15	≤30	≤45	≤60	>60
By Mothers	9	16	21	6	4	2
By Fathers	45	6	6	-	-	-

N = 58 Children

Mean time spent by mothers on each child =

22.88 mins ; S.D. = 20.4

Mean time spent by fathers on each child =

3.36 mins ; S.D. = 7.3

The reported willingness of fathers to participate in the children's treatment does not seem to be reflected in their routine contribution to care. It is perhaps worth drawing attention to the fact that eight of these families, by having two C.F. children, have twice this time commitment daily.

The time involvement tabulated in the Table above is, perhaps, not very impressive, but when the time reportedly spent on each child was compared with the time prescribed for physiotherapy for that child, it was apparent that in 29 of the 58 cases insufficient time was being spent on physiotherapy each day. The most significant factor distinguishing the families who spent the prescribed time on treatment from those who did not, was simply family size

($X^2 = 13.1$, d.f. = 6, $P < 0.05$). This finding was endorsed by the verbal reports of mothers. Those with larger families, and particularly mothers of young children, were likely to find it more difficult to give the C.F. child treatment of the duration prescribed. The mothers of twenty of the children said they found that they resented the time commitment involved in treatment.

On the whole, all but two of the 49 mothers interviewed felt fairly confident, on their own admission, in their handling of the child and his treatment at home. This included all of the mothers who were, on average, providing less treatment than had been prescribed. Many of these mothers were aware of the deficit, and trusted their own judgment to let them know when the child's health required them to make an additional effort over his treatment.

5. Agents of Help.

Because the aim of the study was not only to identify problems, but also, where problems were found, to suggest possible solutions, some consideration was given to the effectiveness of the available social services in meeting the needs expressed by these families.

Three kinds of needs were explored. Needs for help with the ill child and his treatment, for help in the running of the home and for help in babysitting were recorded from mothers' responses during the first interview (Q63 - 65, Table III.B.(xxxi)).

Table III.B.(xxxi) Needs Expressed by Mothers of C.F. Children

Need for help with the child and his treatment	30%
Need for help in running the home	30%
Need for babysitting help	8%
	N = 50 families

The need for baby-sitting help although very important to those who expressed it, did not seem to describe a major problem for the majority of the sample.

The question of help in the home had more widespread relevance. Although 10% of the mothers did have home help, a further 30% felt they required some assistance at home. This need was usually qualified by the explanation that having domestic help would enable the mother to spend more time on the treatment of her C.F. child and, at the same time, would free her from the feeling that by spending time in this way she was neglecting her home. Not surprisingly then, this need was more likely to be expressed by mothers of larger families i.e. more than two children ($X^2 = 5.35$, d.f. = 1, $P < 0.05$). There was a tendency, not reaching statistical significance, for mothers of C.F. children who were not yet at school i.e. under five years of age, also to be more likely to need domestic help ($X^2 = 2.38$, d.f. = 1, $P \simeq 0.20$).

Before considering parents' need for help in the care of their cystic fibrosis children, it was of interest to consider parents' experience of the social service agencies already in existence to help to meet such needs as theirs.

68% of the mothers had had no contact with a Health Visitor from the local authority's clinic and, of those who had had visits from the Health Visitor, 25% had found them most unhelpful. Only 12 of our 50 mothers had then been helped in some way by such visits. However, as a rule these visits terminated when the child went to school, and sometimes earlier.

There had been a marked absence of other visitors to the homes of these cystic fibrosis children, and such visits as were reported by 12 of the mothers were isolated events.

26% of the mothers had been helped by the hospital social worker, although help by this means was usually sought by the mothers during visits to the hospital and was not very readily available.

Clearly then there was no reliable source of help available to assist these families in the home care of their C.F. children. However it was necessary to consider more specifically just what these mothers required. Although, undoubtedly, all of these mothers who expressed a need for help with the child's treatment were referring to their need for assistance in giving physiotherapy, there was the feeling that practical needs were not the only issue, and that underlying needs were perhaps being overlooked.

It was interesting then to invite mothers views on the kind of visits which they would find most helpful, introducing two hitherto ignored topics of professional advice or information and of emotional support. 78% of the mothers felt it would be helpful to have some kind of home visiting service for families affected by cystic fibrosis, although their views on the form

this should take did vary. Table III.B.(xxxii).

Table III.B. (xxxii) Requirements of Home Visits expressed by C.F. Mothers.

Available only on request to give advice on special problems	24%
Available at any time to provide information and moral support	26%
Available to provide advice, information and support	28%
N = 50 families	

Perhaps surprisingly, when the real possibility of the hospital's provision of some such service was raised, only 62% of the mothers welcomed the suggestion wholeheartedly. Two mothers expressed some doubts and the remaining 17 mothers did not approve of the idea. Nevertheless, many of the mothers had described a number of difficulties during the early years of coping with cystic fibrosis, and these observations and the findings reported above, were instrumental in the introduction of a home visiting service as an extension of the hospital's responsibility for the welfare of these families.

6. Hospitalisation

To conclude this chapter summarising the findings most relevant to the medical management of cases of cystic fibrosis,

the children's experiences of hospitalisation must be included. Without setting out especially to explore this aspect, this study can add little to the knowledge already available from good general studies of children in hospital e.g. Robertson (1958). However, hospitalisation looms very large in the lives of several of these children so that the question could not be ignored altogether. The findings are again reported in relation to all 58 of the children in the sample.

The number of times that any one child in the sample had been admitted to hospital ranged from 0 to 10 separate occasions and the mean number of admissions over the sample as a whole was 3. (S.D. = 2.3). The number of weeks spent in hospital by any one child varied from 0 to 50 and showed a mean of 9 weeks. (S.D. = 11.2). Clearly, then, in this small sample, the tremendous variations shown in the hospital experience, even among children of approximately the same age, were such that it was impossible to make valid generalisations.

Varying periods of time had elapsed, between the most recent hospital admission of the child and the time of the enquiry, in the 50 families but it was still felt to be appropriate to ask about the extent to which the child had been prepared for his hospitalisation and whether his in-patient experiences were ever discussed with him. Three of the children had never been in-patients and 23 of the others had been admitted to hospital at an age when it was not possible to prepare them for what was to happen. For the remaining 32 children these were relevant questions.

Just over half of them had been prepared, by their parents, for their admission to hospital, the remaining 14 had not. To some extent, this was a function of the clinic practice whereby children might occasionally be referred directly to the ward from their routine clinic visit, for admission on the same afternoon. It can be argued that when the child's health requires such urgent attention both the mother and the child often suspect that this will happen. Some mothers were glad of this practice which relieved them of the necessity to prepare the child.

Not surprisingly, mothers of children, of whatever age, who try to prepare their children for hospital admission, are more likely to discuss the hospital experiences and procedures with the child afterwards ($X^2 = 34.2$, d.f. = 4, $P < 0.001$).

The children's protests during their hospitalisation were assessed by a symptom count scored from mothers' reports. A number of symptoms of distress exhibited by children were listed and mothers were asked to identify the behaviour(s) which had exemplified their child's response to hospitalisation. (Q.204) Each positive response added one point to the child's hospital protest index. Table III.B.(xxxiii)

Table III.B.(xxxiii) Hospital Protest Index for C.F. Children

Q.204.	In hospital, did he tend to:	Score
	Cry excessively?	1
	Cling to you?	1
	Withdraw from you?	1

Refuse to speak to you?	1
Become very passive and listless?	1
Become very angry with you?	1
Become very angry with his brothers and sisters?	1
Suck his thumb?	1
Rock or bang his head on the bed?	1
Index takes values 0 - 9	

Scores on this index were, perhaps surprisingly, rather low, ranging only from 0 to 6. The mean protest score was only 1.3 and its standard deviation 1.4. The extent of the children's protests seemed to reflect most strongly, their years ($X^2 = 10.1$, d.f. = 4, $P < 0.05$) and their previous experience of hospitalisation ($X^2 = 9.88$, d.f. = 4, $P < 0.05$) but both of these relationships will require some further discussion. Preparation of the child beforehand and even frequent visits from parents seemed to have little effect on the degree of the protest shown.

Behaviour problems, particularly of clinging and of attention-seeking behaviour were fairly common on the children's discharge from hospital and mothers of 22 of the children reported such difficulties when the children returned home. Younger children, ($X^2 = 4.79$, d.f. = 2, $P < 0.10$), and those who had shown most protest against being in hospital, ($X^2 = 8.05$, d.f. = 2, $P < 0.02$), were most likely to present behaviour problems of this kind on their homecoming.

The only other issue which could usefully be encompassed

by the limited scope of the study in this field, was that of parents' attitudes to the in-patient care of their C.F. children. These attitudes were assessed from mothers' responses to questions about the child's response to hospitalisation and about the changes which they would like to see introduced, in the hospital arrangements, to benefit their child.

Some of the views expressed could be readily summarised as showing negatively or positively toned attitudes. The attitudes of parents who described their reservations about an otherwise satisfactory system were described as 'mixed feelings'. Several parents remained non-committal in their views and these were subsumed under the heading, 'no comment'. Table III.B.(xxxiv)

Table III.B.(xxxiv) Parents' Attitudes to the Hospital Experiences of their C.F. Children

No comment	36%
Positive attitude	42%
Mixed feelings	4%
Negative attitude	18%

(Responses from parents of 55 C.F. children who have been in hospital)

Parents' attitudes to their children's hospital experiences did not seem to be related to their attitudes to the hospital

doctors (X^2 = 0.66, d.f. = 2, P $\simeq$ 0.70) although there did seem to be an association between their satisfaction with the way in which the diagnosis had been managed initially and these attitudes to the child's subsequent hospitalisations (X^2 = 3.49, d.f. = 1, P $<$ 0.10).

Having described the family setting into which these C.F. children have been born and having considered the medical background of the diagnosis and treatment of their disease, it is appropriate for us now to examine the impact which these children with cystic fibrosis have on the functioning of their respective families.

C. The Impact of Cystic Fibrosis on Family Functioning

That cystic fibrosis should have an impact on family functioning might have been predicted from the importance attached, in recent literature, to the inclusion of some consideration of the family, in the study of any child. That this disease does indeed intrude in family life has also been suggested by the findings of research specifically involving families affected by cystic fibrosis. However, the nature of the health service in the countries where this previous research has been carried out has made it difficult to separate the impact of the disease itself, from that of the consequent economic stresses and strains. Thus it was important for this study to examine the effect of cystic fibrosis on families within the National Health Service in Britain.

Three areas of family life, in which cystic fibrosis might be anticipated to intervene, were investigated in this study:

1) Financial Pressures
2) Social isolation
3) Family relationships

Information about these three aspects was gathered throughout the interviews with parents, but, towards the end of the second interview with mothers and of the interview with fathers, the parents were specifically invited to comment on the extent to which they felt the child's illness had intruded into family life. Their views on this explicit point are included at the end of this chapter.

1. Financial Pressures

Housing, an important aspect of the material welfare of these families, has already been discussed (Chapter A). The question of financial pressures, specifically in relation to cystic fibrosis, was introduced by the discussion of the difficulties of financing transport to take the child to the out-patient clinic and mothers' responses to these questions were recorded in Chapter B.

With these aspects of possible economic difficulty already clarified, mothers were asked whether they felt the child's illness had placed any other additional strains on the family budget (Q.82). 36% of mothers reported some additional expenses because of cystic fibrosis.

Two areas of expenditure, in which the C.F. child's needs might be found costly, were explored in more detail: food and clothing. Although none of the children was on a special diet, 34% of mothers said that they bought special food for this child. Only one third of them, however, said that this was a source of extra expenditure.

22% of the mothers said they bought better quality, or more, clothing for this child than they would normally buy, although three of them denied that this strained the family budget.

The results indicated that financial strain tended to be experienced most severely by the lower income families who particularly mentioned clinic and hospital visits, and the provision of adequate food and clothing, as sources of real economic difficulty. The financial strain experienced by the families of more adequate means was occasioned by the family trying to provide a higher standard of living for the C.F. child than they could maintain

for the whole family.

The amount of money spent by the mothers in these families, on food alone, ranged from £3-20 per week, with the average amount for housekeeping being £10:50p per week. (S.D. 3.68) However only 48% of the mothers considered this to be enough. Estimates of the additional sum required to allow the mothers to feed their families adequately ranged from £3-10 p.w. and showed a mean requirement of an additional £3.30.

Thus, although only 36% had reported additional expenses because of cystic fibrosis, 52% felt they required more money than they had, to feed their families adequately. It is important to note that these figures were gathered between July, 1971 and June, 1972 before Britain's entry into the European Economic Community and before the recent dramatic increase in the cost of living.

It was perhaps surprising that the financial difficulties reported did not seem to be influenced by whether the mothers had a source of paid employment ($X^2 = 2.3$, d.f. = 3, $P \simeq 0.50$). However, in the terms of this thesis, there was a highly significant relationship between the extent of the financial pressure on the family and the mother's general ability to cope with the situation. The derivation of an Index of Coping has already been hinted at, but full explanation is more appropriate in the context of the next chapter. Here then, given simply that some mothers will be better able than others to cope with having a cystic fibrosis child in their family, we can say that the greater the financial pressure on the family, the less likely it becomes that the mother will be able to cope with the other aspects of her child's disease ($X^2 = 7.49$, d.f. = 1, $P < 0.001$).

Further information about the families' material assets was gathered in terms of the percentages of them who owned washing machines, cars and telephones. Although to some extent ownership of these items may be interpreted as an index of affluence, the need of cystic fibrosis families for these commodities may be judged to be greater than average. Thus the information contained in Table III.C(i) may be of two-edged interest.

Table III.C.(i) Ownership of Household Luxuries among C.F. Families.

Washing machines	84%
Cars	46%
Telephones	32%
from N = 50 households	

Although these figures require comparison with some standard, either in the control group or in the general population, to give them meaning, it is interesting to observe that the ownership of these items among cystic fibrosis families is still primarily a matter of the socio-economic status of the family in question. The higher the family on the socio-economic scale, the more likely they were to possess cars and telephones. Table III.C.(ii). This in itself is not surprising, but, the fact that the ownership of these things was not, at the time of the study, related to the severity of the condition of the child in the family, may suggest a source of financial pressure i.e.

families who might not otherwise have done so, may feel pressure upon them to possess a car or a telephone for the sake of their C.F. child, particularly if his health is of grade C on our scale.

Table III.C.(ii) Association between Material Possessions and Socioeconomic Status, and the Health of the C.F. Child

	Socio-economic Status	Health of Child
Car ownership	$X^2 = 11.00$, d.f. $= 5$, $P < 0.10$	$\chi^2 = 1.31$, d.f. $= 2$, $P \simeq 0.50$
Telephone ownership	$X^2 = 8.35$, d.f. $= 5$, $P \simeq 0.20$	$\chi^2 = 0.37$, df $= 2$, $P \simeq 0.80$
	from N = 50 families	

Financial pressures on a family can be brought about not only by the family's expenditure but also by a loss of income. This possibility was explored within this sample and 13 parents, 12 fathers and 1 mother, were found to have lost work for reasons directly involving cystic fibrosis, and 10 of them had suffered a loss of income as a result. 9 of these parents reported that absences from work, for these reasons, had occurred on several occasions.

Thus the question of financial pressures on families affected by cystic fibrosis was not eliminated entirely by the selection of National Health Service patients.

2. Social Isolation

Financial pressures and social issues overlap in the matter of family outings and holidays. Table III.C.(iii).

Table III.C.(iii). Family Outings and Holidays among C.F. Families

	Outings	Holidays
Family does have:	64%	40%
Family does not have:	36%	60%
Reasons:		
Other than C.F.	28%	40%
".." expense of C.F.	8%	8%
".." worry/inconvenience of C.F.	-	12%
	from N = 50 families	

Factors relating specifically to cystic fibrosis are clearly not the only restrictions on the activities of these families, and considerations of the size of the family and the ages of the children were important intervening variables. In general, a total of 28% of the mothers in the sample felt their family's activities to have been restricted in some way by the cost of the child's illness. Mothers' responses to this question did, however, show highly significant relationships to their ability to cope with the situation and to their own marital difficulties. Thus a mother who was, in our terms, coping well was less likely to complain of the restrictions imposed on the family by the cost

of the child's illness (X^2 = 6.75, d.f. = 1, P < 0.01) as was a mother whose relationship with her spouse was secure (X^2 = 15.89, d.f. = 1, P < 0.001).

Turk (1964) was the first to suggest that cystic fibrosis acted to reduce the social contacts of affected families with their extended family and with their friends, and the notion of social isolation as a concomitant of cystic fibrosis has persisted. To examine the generality of this premise for this sample, mothers were asked in some detail about the frequency of their contacts with family and friends, and whether they felt that the C.F. child had affected their ability to maintain these relationships. Table III.C.(iv).

Table III.C.(iv) C.F. Families' Contacts with Family and Friends

Frequency of Contact	Family	Friends
Daily	26%	40%
At least weekly	34%	28%
At least monthly	12%	16%
Rarely	26%	4%
Never/no family, friends	2%	12%
Frequency of Contact reduced by C.F.	20%	20%
	N = 50 families	

There was no significant evidence of a relationship between mothers' declarations that cystic fibrosis was intrusive and the frequency with which she met her family or friends (Family: X^2 = 2.83, d.f. = 4, P ≃ 0.70; Friends: X^2 = 6.40, d.f. = 4, P ≃ 0.20).

There were clear social class differences in the amount of contact between families and their extended families, with families in social classes I and II being less likely to have frequent contact with their relatives than families lower down the social scale ($X^2 = 6.29$, d.f. = 1, $P < 0.02$). The same was not true however, of contacts with friends, where no significant social class differences were found ($X^2 = 1.17$, d.f. = 1, $P \simeq 0.30$). The time which the family had spent living in its present neighbourhood was not a significant factor in the frequency of contact reported with family or friends (Family : $X^2 = 0.37$, d.f. = 1, $P \simeq 0.50$; Friends: $X^2 = 0.90$, d.f. = 1, $P \simeq 0.30$). Not surprisingly though, mothers who did have more frequent contact with their friends were more likely to report a positive attitude to the neighbourhood in which they lived ($X^2 = 14.56$, d.f. = 1, $P < 0.001$).

These social contacts provided important sources of practical help and emotional support to the mothers and the more frequent the contact, the more likely it was to be advantageous to mothers, as shown by Table III.X.(v).

Table III.C.(v) The Degree of Association between Frequency of Contact and Helpfulness of the Family and Friends of C.F. Families.

	Family	Friends
Frequency of contact x provision of practical help	$X^2 = 15.1$, d.f. = 4 $P < 0.01$	$X^2 = 18.6$, d.f. = 4 $P < 0.001$
Frequency of contact x provision of emotional support	$X^2 = 13.2$, d.f. = 4 $P < 0.02$	$X^2 = 26.2$, d.f. = 4 $P < 0.001$
	from N = 50 families	

Conversely then, social isolation deprives families of these sources of support. Although it must be acknowledged realistically that the children who are most severely affected by cystic fibrosis, i.e. those who are graded C on our scale, may ineed be more likely to cut down their families' social activities, ($X^2 = 8.70$, d.f. = 2, $P < 0.02$). It should also be noted that mothers who scored highly on the Taylor Manifest Anxiety Scale were also more likely to be among those who reported reduced contact with their families ($X^2 = 5.75$, d.f. = 1, $P < 0.02$).

Although the implication has been that social contacts provide a means of help for families, particularly for mothers, the frequency of the contact of itself showed no significant relationship to mothers' ability to cope, (Contact with family x coping, $X^2 = 0.23$, d.f. = 1, $P \simeq 0.70$; Contact with friends x coping, $X^2 = 0.61$, d.f. = 1 $P \simeq 0.50$) suggesting perhaps, that the mothers' coping ability draws more from their own resources than from those of their families.

To summarise the social conditions of these cystic fibrosis families, an index was derived from the information discussed to date. This index did not use the formal socio-economic status values already assigned to the families but it gathered together the findings about the housing, social and financial conditions reported by mothers into a simple 6 point scale. For each of the conditions included in Table III.C.(vi) which described the family in question, a point was added to the scale.

Table III.C.(vi) To derive a simple Index of the Social Conditions of the C.F. families in the Sample (data from the first interview with mothers)

Abnormal marital status of parents e.g. separated (Q15 + Q18)
Father often absent from the home i.e. more than the working day (Q15)
Inadequate housing conditions (scale value 08, from Q27)
Contact with family <u>and</u> friends reduced by C.F. (Q52 + Q62)
Home distant from treatment centre for C.F. child ($\geq$ 15 miles, Q77)
C.F. strains the family budget (Q82)
Index takes values 0 - 6

Although the use of indices leads to a loss of information, this index has been introduced simply as a supplement to the fuller assessment of the situation of these families. It was

introduced to provide a single, simple means of comparing these families with their control group, and, indeed the report of the scores obtained is hard to evaluate without such a reference group. For future reference, then, the mothers of these cystic fibrosis children described their families' social conditions in terms which scored from 0 to 5 on the scale. The mean score on this index was 1.7 and the standard deviation was 1.2.

3. Family Relationships.

In addition to its influence on the frequency of the families' contacts with others, it was suspected that cystic fibrosis might also intrude into the relationships with these others. Mothers were asked whether they felt that their C.F. children were treated in any special way, by relatives or friends, because of their illness. Their responses to questions 274 to 276 on the first schedule were assessed and the influence of cystic fibrosis in the reactions of others to the child was scored as 'marked', 'moderate', 'slight' or 'nil' on the basis of the extent and degree of the concessions made to the child because of his illness. Table III.C.(vii).

Table III.C.(vii) The Influence of C.F. in Relationships with Others

None	23
Slight	8
Moderate	14
Marked	13
N = 58 C.F. Children	

These changes in the reactions of people outside the family to the C.F. child did not seem to be associated with the child's age (X^2 = 2.44, d.f. = 3, P $\simeq$ 0.50). More severely affected children were no more likely to evoke such attitude changes (X^2 = 2.72, d.f. = 2, P $\simeq$ 0.30), although there was evidence of a trend for these changes to be more likely to occur when the family's contact with the people in question had been reduced by cystic fibrosis. This trend reached the 2% level of significance for both relatives and friends of the family. (Reduced contact with the extended family x relationship influenced by C.F.: X^2 = 8.96, d.f. = 2, Friends, X^2 = 7.91, d.f. = 2) Not surprisingly perhaps, there was a strong association then between the influence which cystic fibrosis exerted in relationships with others and the reactions of the healthy children in the family to the situation. Resentment of the C.F. child by his well brothers and sisters was the more likely the more that child's illness intruded in his relationships with these significant others (X^2 = 5.15, d.f. = 1, P $<$ 0.05).

Cystic fibrosis, as we have already observed, has been reported to isolate affected families from those around them. In addition to this, however, the literature describes a degree of isolation of individuals within the family, brought about by the disruptive effect which cystic fibrosis is said to have on family communications. This aspect of the impact of the disease is said to create 'a web of silence' around the child and is held to be an important factor in the emotional development of the affected child. As a precursor then, to the study of the emotional development of these children, the influence of cystic

fibrosis on the network of family communications was explored (Q214-220, mothers' first schedule; Q40-46, fathers' schedule).

Throughout this area of the study, communications were described as 'good', 'moderate', 'poor' or 'nil' on the basis of a scheme which is explained in Table III.C.(viii).

Table III.C.(viii) Assessment Scheme for Family Communications

Good	:	All topics, including cystic fibrosis, are freely raised between the discussants
Moderate	:	Difficulty is experienced in discussing cystic fibrosis but communication is otherwise good
Poor	:	Difficulty is experienced in discussing cystic fibrosis but communication on most topics is impoverished
Nil	:	This refers to relationships with children who are too young for the assessment scheme above to be of relevance to them. It also refers to the missing cases who were not available for interview

To summarise the freedom of exchange of views within the family, four communication channels were considered: that between the spouses; between parent and affected child; between parent and others outside the family and between the child and others. Although information was gathered about communication with a

number of 'outside others' it was considered that the optimal evidence available of communication with one of those 'others' would be sufficient to make the point.

Retrospective information, about communication between parents who had since separated, was entered into the scheme with the rest of parents' responses which are scored in Table III.C.(ix).

Table III.C.(ix) Ease of communication in C.F. families

		Good	Moderate	Poor	None
Mothers' views					
Mother-Spouse	(N=50)	48%	22%	28%	2%
Mother-Others	(N=50)	42%	36%	20%	2%
Mother-C.F.Child	(N=58)	21%	38%	24%	17%
C.F.Child-Mother	(N=58)	28%	38%	19%	15%
C.F.Child-Others	(N=58)	16%	45%	22%	17%
Fathers' views					
Father-Spouse	(N=50)	52%	32%	10%	6%
Father-Others	(N=50)	48%	38%	8%	6%
Father-C.F.Child	(N=58)	21%	34%	24%	21%
C.F.Child-Father	(N=58)	26%	32%	21%	21%
C.F.Child-Others	(N=58)	26%	29%	24%	21%

The influence of cystic fibrosis on family communications is, in these terms, most clearly represented by the 'moderate' column of the above Table III.C.(ix). This signifies otherwise good family communications disrupted by the difficulties which they experience in discussing the very salient topic of cystic fibrosis. Although not, perhaps, as widespread as the literature suggests, communication difficulties are not uncommon, and, where they are effective, they seem to influence more than one line of communication. In general, the probability that difficulty would be experienced in communication in one of the channels, given that difficulty was reported in communication with another family member, was not high. (e.g. mother-spouse x mother-child, $X^2 = 0.25$, d.f. = 1, $P \simeq 0.70$). The exception to this finding was the case of mothers' communication with others outside the family which seemed to be facilitated by their having achieved a 'good' level of communication with their husbands (Mother-spouse x mother-other, $X^2 = 9.34$, d.f. = 4, $P < 0.1$) to a degree which almost reached statistical significance.

Although this factor of family communications is one which will appear repeatedly in interplay with other major factors in this study,it is useful here to consider some of the factors which may contribute to this differential in communicative ability between families.

Although there was a strong relationship between the ease of discussion between spouses as reported by the separate partners ($X^2 = 13.5$, d.f. = 1, $P < 0.001$) the factors significant to the communicativeness of each, were different. For mothers, a better understanding of cystic fibrosis ($X^2 = 3.84$, d.f. = 1, $P < 0.05$)

and a higher degree of satisfaction with the way in which the diagnosis had been communicated to them ($X^2 = 6.73$, d.f. = 1, $P < 0.01$) seemed important precursors of their ability to discuss cystic fibrosis. For fathers, on the other hand, personality factors remained the major determinants of communicativeness. More anxious fathers, as identified by above-average scores on Taylor's scale, were less likely to be good communicators ($X^2 = 4.51$, d.f. = 1, $P < 0.05$). Similarly, more taciturn and introspective fathers, as defined by low scores on Cattell's Factor F of desurgency - surgency, were also more likely to be poor communicators ($X^2 = 4.30$, d.f. = 1, $P < 0.05$). Clearly then the whole question of family communication has to be approached with caution. As the findings are amassed to complete the picture of cystic fibrosis children and their families and as they are evaluated by reference to the control group, so then may the importance of the factor of family communications be clarified.

4. Parents' Views of the impact of Cystic Fibrosis on family functioning.

On the whole, 28% of the mothers and 40% of the fathers of the families in this sample felt that cystic fibrosis had had some impact on the life of their family. There was a significant degree of agreement between parents on this point ($X^2 = 4.24$, d.f. = 1, $P < 0.05$), on the basis of which we can safely say that more than half of the sample do not find cystic fibrosis intrusive in family life. 12% of mothers and 10% of fathers found that cystic fibrosis made it harder for them to be the kind

of parents they wanted to be and again there was a significant degree of agreement between parents ($X^2 = 13.89$, d.f. = 1, $P < 0.001$).

Parents were asked about their experience of a number of specific hardships which might have been imposed by the influence of cystic fibrosis. These hardships related primarily to time, money and the quality of life in general. (Q.41 of mothers' second schedule; Q.95, fathers' schedule). For each positive response a point was added to a total which ranged from 0 - 7 for mothers and from 0 - 9 for fathers. Parents were also asked which of all of these hardships was the most serious and the rank ordering of these hardships which resulted from their cumulated responses is given, with the distribution of total scores, in Table III.C.(x).

The findings of this chapter would seem to indicate that cystic fibrosis does indeed have some impact on the financial, social and interpersonal aspects of family life, but, that the effects are not as dramatic as those described by earlier studies. The last table of this chapter suggests the hypothesis that the parents of the children may take the brunt of the effect of cystic fibrosis. Information gathered from the study to describe the influence of the disease on the personal lives of the parents of affected children is presented in the next chapter.

Table III.C.(x). Family Hardships occasioned by Cystic Fibrosis (in order of decreasing severity)

Mothers' views of most serious hardship	
No hardship	15
Less fun in life	12
Less spare time	10
Less money for mother to spend on herself	8
Less energy (mother)	2
Less time to play with the children	1
Less time alone with husband	1
Less money to spend on the other children	1
	N = 49 mothers

Mothers' Total Scores over all seven hardships ranged 0 - 7

Mean: 2.2, S.D. = 2.2

Fathers' views of most serious hardship	
No hardship	24
Less fun in life	11
Less money to spend on self	5
Less spare time	4
Less time alone with wife	2
Less energy	1
Less money to spend on wife	0
Less money to spend on the other children	
Less money to spend on the house	
Less time to play with the children	
	N = 47 fathers

Fathers' Total Scores over all nine hardships ranged 0 - 8

Mean: 1.9, S.D. = 0.4

D. The Effect of Cystic Fibrosis on Parents.

It is well known that the first questions asked, by most mothers, after giving birth, is "is he/she all right?" The relief that comes from a positive response to this question suggests that mothers themselves anticipate dire consequences if the child is not "all right". If the child is not normal, for instance, if he has cystic fibrosis, the literature leads us to expect, among these dire consequences, parental rejection of the child, marital strife and deterioration in the mother's physical and mental health. However, these gloomy predictions have been based on the findings of rather narrow studies of small numbers of mothers and may not be representative of the situation in this sample. To simplify our assessment of the effect of cystic fibrosis on the lives of the parents of the children in this sample, three facets of the parents' lives were considered separately:

1) The Parent as a Parent of a C.F. Child

2) The Parent as a Spouse

3) The Parent as an Individual

Although fathers, on the whole, spend less time in their parental roles than do mothers, their attitudes to parenthood and to their children, are nonetheless, of considerable importance to their families. Thus, in this chapter, parallel views, from both parents have been recorded as far as possible. Towards the end of the chapter an index of maternal coping ability is derived. This serves the dual purpose of summarising the difficulties raised for mothers of C.F. children and of assessing the extent to which they are able to deal with these problems.

1. The Parent as a Parent of a C.F. Child

To the normal responsibilities of parenthood, further commitments are added by cystic fibrosis, none are taken away. In order then, to assess the influence of the disease upon the parents, both aspects of their parental role have to be considered i.e. caring for a chronically ill child and meeting the normal needs of a growing child.

Parents' attitudes to the child and his illness were recorded in some detail during the interviews with fathers, and during the second interviews with mothers, since it was felt that it would be in these attitudes that the effect of the disease on the parents would be revealed.

Fathers seemed more inclined to doubt the diagnosis than were their wives (Table III.D.(i)) and although there was a high degree of agreement on this point between parents ($X^2 = 7.59$, d.f. = 1, $P < 0.01$) it was interesting to observe the marked difference, between parents, in the factors associated with such doubt. For mothers, who do spend a lot of time in the care of these children, the most significant factor associated with the incidence of doubt among them, was the realistic one, of the number of the child's symptoms ($X^2 = 8.45$, d.f. = 1, $P < 0.01$). The severity of the child's condition, as clinically assessed, was an important factor too, but one which did not reach statistical significance in its relationship with the occurrence of doubt in mothers' minds ($X^2 = 7.59$, d.f. = 4, $P \simeq 0.10$). For fathers, there were no clear-cut factors which seemed to be related although there was a trend, not reaching statistical significance, for the severity of the child's physical condition

to be influential (X^2 = 3.30, d.f. = 1, P < 0.10). A relationship of comparable significance was detected between expressed doubt and the fathers' anxiety as measured, suggesting that in some cases this doubt may be acting as a defence mechanism against the unpleasant truth (X^2 = 3.26, d.f. = 1 P < 0.10).

Table III.D.(i). Parents' Reactions to their Child's Illness - 1. Doubts.

	Mothers	Fathers
Parent doubts the diagnosis	26%	40%
Parent is willing/has been willing to experiment with the child's treatment	40%	18%
	N = 50 families	

To a considerable extent parents have to monitor their child's treatment and may have to introduce minor variations in his routine on the basis of their own judgment e.g. dosage of substitute pancreatic enzyme is varied according to the child's diet and the nature of his stools. However it was feared that experimentation might go further than this, particularly when the incidence of doubt of the diagnosis was high. Surprisingly, this did not prove to be the case. Although 40% of the mothers did admit to having experimented with their child's treatment at some

stage, "just to see if he really needs it," this admission did not seem to be related to the incidence of doubt among the mothers ($X^2 = 0.74$, d.f. = 1, $P \simeq 0.50$). Fathers were less willing to experiment with the child's treatment and verbal evidence suggests they were not always aware that their wives had done so. (Table III.D(i)).

Physiotherapy was the aspect of treatment most subject to this experimentation. We have already observed that in a number of cases the time spent in this treatment was rather less than that prescribed. Now, we find that in a number of families it has, at some stage,been omitted altogether. It must however be stressed that these experiments with treatment occurred only occasionally and were usually of short duration in the life of any one child.

Objectively, a burden is certainly placed upon all these parents but the extent to which it is subjectively perceived as such, by the parents themselves, is another matter. Parents were asked whether they ever felt the burden of caring for a C.F. child was too great. (Q.20, mothers second schedule; Q.81, fathers' schedule). Not surprisingly, this question was more relevant to mothers' experience and the fathers who gave positive responses to this question qualified them by explaining that the brunt of the burden was borne by their wives. Table III.D.(ii).

Table III.D.(ii). Parents' Reactions to their Child's Illness - 2. re Burden of C.F.

	Mothers	Fathers
Parent has felt the burden of C.F. to be too great	40%	24%
Parent has felt unable to fulfil other responsibilities	44%	18%
	N = 50 families	

Although fathers who had experienced this feeling of being too heavily burdened had attributed much of this burden to their wives, there was not a high degree of agreement between parents' reports on this point ($X^2 = 0.77$, d.f. = 1, $P \simeq 0.50$). There was a much stronger probability that those fathers who reported finding cystic fibrosis a burden would also be those fathers who had found that the child's illness had influenced or interfered with their work in some way ($X^2 = 3.92$, d.f. = 1, $P < 0.05$). For mothers, the feelings of being burdened did not relate to family size or indeed to the severity of the child's condition but mothers living in poor social conditions, as gauged by our index derived in sub-section III A, were significantly more likely to report such feelings ($X^2 = 5.97$, d.f. = 1, $P < 0.02$). There was a trend, not reaching statistical significance, for

mothers who had reported that their husbands helped them in their homes, to be less likely to feel burdened in this way ($X^2 = 3.15$, d.f. = 1, $P < 0.10$).

If parents feel they are labouring under a burden, in caring for their C.F. child, it was reasoned that they might well then feel unable to meet other responsibilities, Table III.D.(ii), and in some cases this proved to be so. However it is only fair to point out that for the 22 mothers for whom this was a source of worry there were only 5 for whom neglect of other responsibilities seemed to be the norm. The remaining 17 mothers, representing 34% of the families in the sample, found this a recurrent rather than a constant problem. Responsibilities to the other children in the family and to husbands were the first to suffer if the C.F. child required more attention. Fathers' views of unfulfilled responsibilities were on a different plane altogether, and the concern they expressed in response to this question was usually for the extended family, for the fathers' own parents, whom he felt he had neglected, in having to meet the needs of his own nuclear family.

Understandably, parents do have periods of feeling very discouraged by cystic fibrosis and it seemed likely from their responses that parents who denied such feelings were drawing heavily on their psychological defence mechanisms. There was concern however, lest the despondency be rooted in feelings of guilt or responsibility for the child's condition and this possibility was put to parents (Q.24 mothers' second schedule; Q85, fathers' schedule). The incidence of positive responses is scored in Table III.D.(iii).

Table III.D.(iii) Parents' Reactions to their Child's Illness - 3. Despondency and Optimism.

	Mothers	Fathers
Parent feels discouraged by C.F. sometimes	68%	48%
Parent feels responsible for child's condition	30%	36%
Parent feels optimistic about child's progress	78%	80%
	N = 50 families	

Although about one third of the parents said they felt responsible for the child's illness, this could not be interpreted as an indication of feelings of guilt in all cases, for several parents had accepted their genetic responsibility as something that had been outwith their control. However, verbal observations made by parents about the way in which the diagnosis had been communicated to them, suggested that the management of this situation could do a great deal to eliminate feelings of guilt before the took root.

In spite of all of the difficulties, it was encouraging to discover that a large majority of the parents were able to maintain a feeling of optimism about the progress the child was making (Table III.D.(iii)), although the optimism expressed was in no way related to the severity of the clinical condition of the

child in question (Mothers: $X^2 = 3.10$, d.f. = 4, $P \simeq 0.50$; Fathers: $X^2 = 3.30$, d.f. = 4, $P \simeq 0.50$) In this light, such optimism may be seen to be misplaced, another manifestation of the ubiquitous system of psychological defences, but, without this optimism or hope, it would be quite impossible for these parents to care for their C.F. children or to tolerate these other feelings which we have just described, which are aroused in them by doing so.

The principles of the social psychology of human groups and the concepts of group therapy suggest that some of the difficulties, both practical and emotional, created for parents by cystic fibrosis, might be alleviated by contact with others who understand and share these problems. Indeed the introduction of parents' groups or at least of mothers' groups among families suffering from other ailments has been reported to have tremendous therapeutic value for these parents. In spite of this, the branch of the C.F. Trust which held meetings for parents in the region covered by this sample,was known, at the time when the study was begun, to be having difficulty in attracting members. Having explored parents' reactions to the illness it is of interest now to consider their attitudes to meeting other parents who share their misfortune and to the concept of a C.F. Parents' Group. Information on this point came primarily from mothers.

We have already observed (Sub-section III.B) that the literature produced by the C.F. Trust to aid parents' understanding of the disease was not widely circulated in the Edinburgh area, although most parents were familiar with its newsletter, and so, presumably, were aware of the existence of the C.F. Research Trust

as an organisation. , Most of the 18% of mothers of families in this sample who had never heard of the Trust were those whose children were not patients at the Royal Hospital for Sick Children. Nevertheless, only 36% of all the mothers said they were members of the parents' group and only one third of the mothers who did not know of these meetings, said they would have gone had they known of them. Furthermore, at the time of the investigation, parents of eightof these member families reported that their attendance at meetings had lapsed, so that only parents from ten families in the whole sample seemed to be regular supporters of the Parents' Group. All of those parents who said they were members of the Parents' Group had been to some meetings ($X^2 = 30.7$, d.f. = 2, $P < 0.001$).

The only really significant factor differentiating Parents' Group members from non-members was social class status ($X^2 = 8.26$, d.f. = 1, $P < 0.01$). Parents of Classes I and II predominated and parents of lower social classes were correspondingly grossly under-represented at these meetings. This finding was endorsed by the views expressed by mothers at interview. Although the issues of geographic location, in terms of distance from the place where meetings were held, and of the age of the child or the size of the family, were all raised as difficulties by parents who did not attend these evening meetings, none of these factors was significant in distinguishing members from non- members Table III.D.(iv.)

Table III.D.(iv) Factors in Membership of the C.F. Parents' Group.

Table III.D.(iv) Factors in Membership of the C.F. Parents' Group.

Number of total sample who were members	18
	Members vs. Non-Members
Social Class	$X^2 = 8.26$, d.f. = 1, $P < 0.01$
Distance from meetings	$X^2 = 7.80$, d.f. = 4, $P < 0.10$
Family size	$X^2 = 9.30$, d.f. = 6, $P \simeq 0.20$
Age of C.F. child ($<$ or $\geqslant$ 5 years)	$X^2 = 0.10$, d.f. = 1, $P \simeq 0.90$
	N = 50 families

Members and non-members also showed some significant personality differences. Those who scored high on Cattell's Factor I, that is, those whom he would describe as tender-minded, dependent and sensitive, were more likely to be among the Parents' Group members ($X^2 = 7.55$, d.f. = 1, $P < 0.01$). There was a trend, not quote reaching statistical significance, for group members to have lower scores on Cattell's factor Q_2 than did non-members. Thus those whom Cattell described as group-dependent were indeed more likely to be members of the Parents' Group ($X^2 = 3.71$, d.f. = 1, $P < 0.10$).

At the time of the enquiry the function of the Parents' Group was not well defined even for its members and very disparate views were expressed about the roles of fund-raising, information dissemination, moral support and social activities within that context. These views are not readily amenable to numerical analysis and are better included in the discussion to follow.

It may be enlightening here to note that when parents attitudes, both to the concept of an association for C.F. parents and to the less formal matter of being able to meet other parents of C.F. children, were coded, as being wholeheartedly positive or otherwise, there was a strong connection between the two. Among members positive attitudes, both to meeting other parents and to the group itself, prevailed, although those who expressed less favourable attitudes to one, were more likely to express unfavourable attitudes to the other (X^2 = 8,40, d.f. = 1, $P < 0.01$). Among non-members, the relationship still was apparent, but the majority of these parents had reservations about meeting other parents of C.F. children in the context of the Group or elsehwere (X^2 = 9.39, d.f. = 1, $P < 0.01$). These findings provide a striking exception to the findings among parents of children with other problems, whose attitudes, as reported in the literature, are usually strongly in favour of group formation.

In the literature referring to the impact of chronic illness on the parents of affected children, great importance has been attached to the question of their 'acceptance' of his condition. Hewett (1970) suggested that this umbrella term involved at least two major adjustments on the part of parents. The first issue, of whether the parents accept that the diagnosis applies to their child, has been discussed in the first half of this chapter and it is to the second major adjustment, to the acceptance of the child's limitations, that we now turn.

The question of over-protection and over-dependence in parent-child relationships has indeed been a vexed one and the desirability of avoiding the need to make value judgments in assessing parents' attitudes was recognised from the outset.

This whole area of interest was introduced to parents in a general way by enquiring about any differences which might have occurred in their handling of their C.F. child, relative to their expectations of normal children. Specific aspects of their child-rearing practice were then explored in some detail and it was only towards the end of this section of the interview that parents were asked about their own assessment of the way in which they were bringing up their C.F. child. These matters were discussed in the terms of questions 252 - 271 of the mothers' first interview schedule and in Q.61 - 73 of the schedules for fathers. Parents expectations for each of the 58 children in the sample were considered separately.

On the whole, the mothers of 23 of these children, and the fathers of 25 of them, reported that they did indeed handle their C.F. children differently from their other children and there was a high degree of agreement between parents on this point ($X^2 = 18.0$, d.f. = 2, $P < 0.001$). Early experiences of the parents were clearly influential in these latter attitudes and parents who had reported a number of difficulties with their child during infancy were significantly more likely to report a change in their attitude to the child at the time of the enquiry (Table III.D.(v)). The way in which the diagnosis had been communicated to parents was also an important factor, since parents reporting a higher degree of satisfaction with the management of this situation were significantly more likely to have altered their expectations of the child in question. Table III.D.(v).

Table III.D.(v). Factors in the Alteration of Parents' Attitudes to the upbringing of their C.F. Children.

	Mothers	Fathers
Altered attitudes to this child	$^{23}/_{58}$	$^{25}/_{58}$
x infant difficulties	$X^2 = 8.98$, d.f. = 1, $P < 0.01$	$X^2 = 18.6$, d.f. = 1, $P < 0.001$
x satisfaction with the management of his diagnosis	$X^2 = 5.30$, d.f. = 1, $P < 0.05$	$X^2 = 2.380$, d.f.= 1, $P < 0.10$
x severity of the child's condition	$X^2 = 1.55$, d.f. = 2, $P \simeq 0.50$	$X^2 = 7.08$, d.f. = 2, $P < 0.05$
	N = 58 children	

Eight aspects of parents' attitudes to their C.F. children were then considered. By scoring one point for a slight difference, two for a moderate difference and three for a marked difference in the parent's attitude to this child, an index, taking values 00-24, could be derived for the extent of change reported in the child-rearing attitudes of each parent. On this basis, the composite scores of mothers ranged from 00-16, showing a mean of 4.5 and a standard deviation of 4.0. Fathers' scores ranged from 00-15 and showed a mean of 4.2 and a standard deviation of 3.9. On the whole these scores reflected parents' responses to the introductory question of this section ($X^2 = 18.3$, d.f.=1, $P<0.001$)

but there were a few parents, five in number, who did not imagine that cystic fibrosis had influenced their handling of their child but who, later went on to discuss specific concessions which had been made to his condition.

Since these issues were couched in terms of parents' expectations it was possible to put all of the questions to all of the parents. This has to be recalled in the interpretation of some of the findings, since some of the questions were thus rather hypothetical to the parents of younger children. Similarly, the implication was that these attitudes to the C.F. child were to be compared with the parents' attitudes to their normal children. Three families in the sample had only one C.F. child and no healthy children, and four of the families having 2 C.F. children had no other well children, so that it was slightly more difficult for these parents to assess the extent to which their attitudes had been changed. Nevertheless it was felt to be preferable that parents should give their own assessments of their child-rearing principles in relation to this child rather than that the investigator should have to make moral judgments of their behaviour as reported.

Considering first the child at home, it was found that less than one fifth of the parents had modified their expectations of the child's behaviour in this setting Table III.D.(vi). There were no sex differences in these concessions to cystic fibrosis nor was their any significant relationship with the severity of the child's condition, although there was a tendency for the oldest children to be more likely to be excused household chores than they would otherwise have been.

Table III.D.(Vi) Parental Expectations of the C.F. Child at Home

	Mothers	Fathers	Agreement Between Parents
Less expected of the child in personal responsibility for clothing, cleanliness or neatness	20%	16%	$X^2 = 14.0$, d.f.=2 $P < 0.001$
Parents' views x child's age	$X^2 = 2.42$, d.f. = 2, $P \triangleq 0.30$		
x child's sex	$X^2 = 0.47$, d.f. = 1, $P \triangleq 0.50$		
x severity of child's condition	$X^2 = 2.17$, d.f. = 2, $P \triangleq 0.30$		
Less expected of the child in responsibility for household chores	20%	20%	$X^2 = 13.6$, d.f.=2 $P < 0.001$
Parents' views x child's age	$X^2 = 14.7$, d.f. = 2, $P < 0.001$		
x child's sex	$X^2 = 0.29$, d.f. = 1, $P \triangleq 0.70$		
x severity of child's condition	$X^2 = 1.64$, d.f. = 2, $P \triangleq 0.50$		

(Information from 49 mothers and 47 fathers re 58 C.F. Children)

For mothers, however, the factor of family size seemed to be important, for there was a trend, approaching statistical significance for mothers of more than two children to be more likely to make these concessions than mothers of only one or two children ($X^2 = 2.78$, d.f. = 1, $P < 0.10$).

Parental expectations of the child's school performance were a different matter. One third of the mothers and 22% of fathers felt that they expected less from this child in the way of school achievement, but there was no significant relationship between the views of parents on this point ($X^2 = 3.45$, d.f. = 2, $P \simeq 0.20$). Only two mothers and one father felt that, on the contrary, they particularly wanted this child to do well at school, by way of compensation for his physical limitations. There was no significant differences between the ages of the children whose parents expected less of them at school and those whose parents did not ($X^2 = 0.06$, d.f. = 1, $P \simeq 0.80$). There were trends, approaching statistical significance, for girls to be more likely to be excused than boys ($X^2 = 2.79$, d.f. = 1, $P < 0.10$) and for those in poorer health to be more likely to be excused than those who were fitter ($X^2 = 4.68$, d.f. = 2, $P < 0.10$).

Parents' expectations of their C.F. children were most commonly altered in relationto the children's activities outwith the home Table III.D.(vii).

Table III.D.(vii). Parental Expectations of the C.F. Child Outwith the Home.

Table III.D.(vii). Parental Expectations of the C.F. Child Outwith the Home.

	Mothers	Fathers	Agreement between Parents
65. Less expected in relationships with other children	26%	26%	$X^2 = 8.45$, d.f. = 2, $P < 0.02$
66. Less independence allowed in decision making	33%	34%	$X^2 = 10.6$, d.f. = 2, $P < 0.01$
67. Less independence allowed in joining activities away from home	38%	45%	$X^2 = 11.5$, d.f. = 2, $P < 0.01$
68. Parent more cautious in allowing child to join in physical activities	34%	39%	$X^2 = 3.47$, d.f. = 2, $P \simeq 0.20$
(Responses from 49 mothers and 47 fathers of 58 children			

None of these aspects of change in parents' attitudes showed any significant age or sex differences and only the diminished expectations of the child in terms of peer relationships

($X^2 = 6.52$, d.f. = 2, $P < 0.05$) showed any significant relationship to the severity of the child's condition.

38% of mothers and 43% of fathers of children in the sample reported that they were less likely to punish these children and their attitudes in this matter again showed no significant relationship to the age or sex of their children nor to the severity of the child's condition.

A search for personality correlates of parents' attitudes to their children, assessed in these terms did not bring to light any factors which distinguished parents whose attitudes towards their children had been modified and those who had not.

From the findings reported here it was apparent that 26% of the mothers and 32% of the fathers of these 58 children had made no concessions to the child's illness. These parents insisted that they were bringing up their C.F. children in accordance with their expectations of normal children and they were quite happy with their child-rearing methods. A further 31% of the mothers and 38% of the fathers admitted making some concessions to that child but were, nevertheless, content with their behaviour and attitudes toward him. The remaining 25 mothers and 16 fathers were suffering some conflict in this matter. They were all making allowances for their C.F. children but they all reported a degree of dissatisfaction with their upbringing of these children.

Before moving on to consider the influence of cystic fibrosis on another aspect of the lives of these parents, this is an appropriate moment to draw attention, briefly, to the participation of fathers in the upbringing of these children.

As might have been anticipated, from the previously reported findings in relation to treatment, fathers' participation in the care of their cystic fibrosis children is considerably less than mothers'. Table III.D.(viii).

Table III.D.(viii) Fathers' Participation in the Care of C.F. Children.

	Never	Rarely	Frequently	Usually
Fathers' attendance at clinics	42%	36%	8%	14%
Fathers' help with home treatment	28%	22%	38%	12%
N = 50 families				

Approximately one third of the fathers, 17 in number, did add however, that they would like to be able to participate more in the treatment of their C.F. children

In more general terms, mothers were asked about the nature of help which they had from their husbands with all of the children(Q.272 - 273 of first interview schedule). Their responses were scored according to whether fathers helped only

by taking the children out to allow mothers to carry on with domestic chores or whether their participation extended beyond the pleasurable activities, to helping with the chores as well. In fact, 57% of the fathers were reported to be willing to help their wives at home, commonly refusing only at nappy-changing. 19% helped only with the children only by taking them out or by playing with them while as many as 24% were said to take no active part in the child's upbringing at all. It will be easier to assess the significance of these findings when the corresponding data is available about the activities of fathers in the control group.

2. The Parent as a Spouse

It is sometimes forgotten, in studies of this kind, that mothers are also wives and fathers also husbands, and that in the study of children and their families some consideration must be given to the marital relationship upon which the family is founded. This study attempted to give a brief account of the influence which parents felt cystic fibrosis had had on their marital relationship (Q.42 - 49, mothers' second interview schedule; Q.96-102, fathers' interview schedule.)

14 of the 49 mothers (29%) and 20 of the 47 fathers (43%) available for interview, did say they felt that the child's disease had created some difficulties for their marriage and there was a significant degree of agreement between parents on this point ($X^2 = 7.24$, d.f. = 1, $P < 0.01$). Happily two of the mothers and seven of the fathers reported that these difficulties had been resolved by the time of the interview. It was important for the

study to attempt to understand the reasons for these difficulties.

It had been anticipated that the general understanding of the inheritance of cystic fibrosis would prove to be imperfect. Parents were therefore asked if they had ever experienced feelings of resentment or anger as the result of a mistaken belief that their child's condition originated solely in the spouse's pedigree. Such blame was found to be unusual, reported by only 4% of the mothers and 9% of the fathers in the sample.

26% of the mothers and 24% of the fathers did feel that the child's condition had disturbed the physical aspect of their relationship and this was a highly significant factor in parents' previous response to the more general question of the influence of the disease in their marriage ($X^2 = 27.2$, d.f. = 1, $P < 0.001$). 16% of the mothers reported a continuing fear of becoming pregnant because of the risk that any further child might be affected by cystic fibrosis. This fear persisted, in spite of the perfectly adequate methods of contraception used. For the remainder of these parents, cystic fibrosis was said to be instrumental in the disruption of their sexual relationship by the problems of physical fatigue and mental preoccupation with the child which it created for these mothers. This was in part borne out by the finding that highly anxious wives, as identified by above average scores on Taylor's scale, were more likely to report difficulties in this aspect of their relationship with their husbands than were less anxious wives ($X^2 = 7.32$, d.f. = 2, $P < 0.05$).

Difficulties in the physical aspects of their relationship were not the only significant factors contributing to parents' experiences of marital tension, at least from the mothers' viewpoint.

Mothers living in poor social conditions, as indexed in the previous chapter, were significantly more likely to complain of marital difficulties ($X^2 = 6.86$, d.f. = 1, $P < 0.01$) and those who reported a lack of help from their husbands in bringing up the children were also more likely to experience tension in their marital relationship ($X^2 = 6.41$, d.f. = 1, $P < 0.02$). Thus, difficulties of this kind cannot be simply explained in terms of any single factor and these findings must again be interpreted with caution.

Since social isolation in relation to the family unit had already been discussed, it was of interest to extend the scope of this question to include the effects of the social activities of the parents. 22% of the fathers and 54% of the mothers had already commented that they did feel that cystic fibrosis had caused them a degree of social isolation. Parents were then asked to estimate the frequency with which they were able to go out socially, together and separately. Table III. D.(ix).

Table III.D.(ix). Estimated Frequency of Social Outings by C.F. Parents.

Table III.D.(ix). Estimated Frequency of Social Outings by C.F. Parents.

	Weekly	Monthly	Rarely	Never
Both parents (mothers' report)	16%	33%	31%	20%
Both parents (fathers' report)	9%	40%	41%	10%
Mother alone (mothers' report)	43%	14%	19%	24%
Mother alone (fathers' report)	43%	16%	31%	10%
Father alone (mothers' report)	59%	16%	16%	9%
Father alone (fathers' report)	74%	4%	18%	4%
N = 49 mothers and 47 fathers				

The frequency of these outings was higher than that reported in the American studies, where the families tended to be described as being highly restricted in their social activities. Predictably though, there was a social class difference in the pattern of these activities with parents higher up the scale being more likely to go out together than parents lower down the scale ($X^2 = 3.84$, d.f. = 1, $P < 0.05$). Mothers who, in the terms of this thesis, were coping adequately with the child and his illness, were also significantly more likely to go out with their husbands than those who did not ($X^2 = 4.56$, d.f. = 1, $P < 0.05$). For parents going out individually, a significant factor seemed to be that of family size and parents of larger families were less likely to go out for their own pleasure than were parents of smaller families i.e. those having only one or two children.

(Mothers, $X^2 = 5.31$, d.f. = 1, $P < 0.05$; fathers, $X^2 = 5.29$, d.f. = 1, $P < 0.05$).

Because it was felt undue weight had, in the past been given to the destructive effects of cystic fibrosis on parents' marital relationships, this section of the study was concluded by asking parents whether they felt that having a child with cystic fibrosis had drawn them together in any way. Among the 50 families in this sample, 42% of the mothers and 34% of the fathers did indeed report that they felt their relationship to have been strengthened by the need to share the problems that had arisen since the birth of their C.F. child.

3. The Parent as an Individual

Previous research has suggested that some of the most marked repercussions of the diagnosis of cystic fibrosis in a child, are to be seen in the physical and mental health of his parents. Since the parents of these children have a fairly demanding role to play in the medical care of their children, as well as in the normal aspects of their upbringing, and since a deterioration in their health might be expected to have a deterious effect on their ability to fulfil these functions, it is important for us to try to understand the factors which are at work, in order to be able to halt the downward spiral of effect that could ensue from such a chain of events.

Mothers'were asked about selected aspects of their general health (Q.1-5, second interview) referring to the time from the birth of their first fibrocystic child until the time of the interview. Appetite disturbances were not common,

reported by only 7 of the mothers but problems of sleeplessness, depression and nervousness showed a high incidence. Table III. D.(x).

Table III.D.(x). Incidence of Health Problems among C.F. Mothers.

	Sufferers	Have sought medical aid
Sleeplessness	79%	12%
"Run down or depressed"	86%	49%
"Nervous"	57%	31%
	Total N = 49 mothers	

If one point is added for each of these complaints, including appetite loss, and a further point is added for each medicament required then a seven point scale of the mothers' complaints can be derived. The mean score obtained by these mothers on this scale was then 3.0.

26 of the 49 mothers in the sample attributed their health problems solely to the anxiety that their child's disease caused them and a further 14 of the mothers held the influence of cystic fibrosis at least partly responsible for their troubles. Mothers were also asked about their health during childhood. Although 11 of them had had some serious illness as children the incidence of ill health in childhood did not show any significant relationship to the reported health problems of the time of the interview ($X^2 = 0.36$, d.f. = 1, $P \triangleq 0.50$). Similarly, 12 of the

mothers had suffered ill health as adults prior to the diagnosis of cystic fibrosis in their family, but again this showed no significant relationship to these more recent complaints. ($X^2 = 0.003$, d.f. = 1, $P \simeq 0.95$). On the other hand, highly anxious mothers, as defined by Taylor's scale, were significantly more likely to report health problems ($X^2 = 5.65$, d.f. = 1, $P < 0.02$) as were mothers of children who were more severely affected by the disease ($X^2 = 5.03$, d.f. = 1, $P < 0.05$).

Whereas only 6 of the 49 mothers had no such problems at the time of the enquiry, only 27 of the 47 fathers interviewed reported any disturbances at all and 18 of them specified reasons other than cystic fibrosis as the source of their problems. Only 5 fathers attributed their troubles to the effects of worry occasioned by cystic fibrosis, while a further 4 fathers felt cystic fibrosis might have been partly responsible for their ill-health. Table III.D.(xi).

Table III.D.(xi) Incidence of Health Problems among C.F. Fathers

	Sufferers	Have sought medical aid
Appetite loss	4%	-
Sleeplessness	15%	6%
'Rundown or depressed'	43%	8%
'Nervous'	19%	11%
	N = 47 fathers	

The mean score obtained by fathers on the cumulated health index was 1.0. Although 17 of the fathers had been ill as children and 14 of them had suffered ill health as adults before the diagnosis of cystic fibrosis in the family, there was no significant relationship between the fathers' histories and the current incidence of ailments (childhood ill-health, $X^2 = 0.97$, d.f. = 1, P $\simeq$ 0.30; adult ill-health, $X^2 = 0.84$, d.f. = 1, P $\simeq$ 0.30). Unlike the mothers the fathers problems did not show a significant relationship to the severity of their children's condition ($X^2 = 0.07$, d.f. = 2, P $\simeq$ 0.98) although like the mothers, the more anxious fathers identified by Taylor's scale were more likely to have health problems ($X^2 = 4.37$, d.f. = 1, $P < 0.05$).

As has already been indicated, some of these parents did indeed obtain high anxiety scores. 45 mothers who completed Taylor's Manifest Anxiety Scale obtained scores which ranged from 3 to 40, showing a mean of 22.4 and a standard deviation of 9.4. The distribution of 41 fathers' scores ranged from 2 to 30 and had a mean of 14.8 and a standard deviation of 8.6. Taylor's own work suggested a distribution of scores from 1 to 36 among samples of adults and young adults, showing a mean of 14.56. The mean score obtained by women was not significantly higher than that obtained by men, in her sample.

In addition to the effect of cystic fibrosis on the health of parents of affected children, some thought was given to the personal frustrations occasioned for these parents as individuals. We have already recorded that a number of fathers lose hours at work for their C.F. children. Of wider ranging implications was

the finding that 6 of the fathers reported that the child's illness had influenced their work in another way. Fathers of these children tended to turn down job opportunities which would involve ~~either~~ uprooting the family to live elsewhere, particularly if the projected location was to be abroad. Jobs which required fathers to go abroad alone or to work away from home for long periods also tended to be avoided and in some cases the fathers thus lost opportunities for better paid jobs. Only three of these fathers expressed any resentment over the sacrifices they had made. In more general terms, 9 of the fathers in the study said they felt their lives' ambitions to have been altered because of their children's illness.

For mothers too, there were personal frustrations: 10 of the mothers would have liked to have taken up outside employment but felt that they had to remain at home for the sake of their C.F. children; 11 mothers felt C.F. interfered with their domestic pursuits and 14 mothers reported that their ambitions in life had been modified by having a child with cystic fibrosis.

Throughout these interviews parents had been given a number of opportunities to express resentment about the difficulties imposed on their lives. Both parents had been invited to express their feelings about their job restrictions, about the intrusions into their family life and into their parental and marital relationships caused by cystic fibrosis. Feelings about the genetic basis of the disease had also been discussed by both parents and the question of time commitment involved in treatment had been raised with mothers. The number

of issues on which parents expressed resentment, from a poossible total of 5 for fathers and 6 for mothers, is shown in Table III.D.(xii).

Table III.D.(xii) Degree of Resentment Expressed by C.F. Parents.

	Mothers	Fathers
None	26	32
Resentful of a single issue	7	11
Resentful of two issues	8	3
Resentful of more than two issues	8	1
	N = 49	N = 47

Since resentment seemed to be stronger among mothers the analysis of the findings, to identify related factors, was conducted only on the basis of their responses. Resentment tended to be stronger among younger mothers ($X^2 = 6.35$, d.f. = 1, $P < 0.02$) and stronger too, among mothers of social classes I and II than among mothers of families lower on the social scale ($X^2 = 6.67$, d.f. = 1, $P < 0.01$). There was a strong association between the extent of the difficulties presented by the child in his pre-school years and the likelihood that his mother would express resentment ($X^2 = 12.9$, d.f. = 1, $P < 0.001$), although it also seemed to be the case that mothers who were better able to cope with the situation were less likely to be resentful ($X^2 = 4.82$, d.f. = 1, $P < 0.05$).

These feelings of resentment were important to our study, not only as a source of discomfort to the mothers but also as a disruptive influence on their relationships with their C.F. children. A hint of the difficulties that might ensue was detected in the mothers descriptions of their C.F. children (Q.328-331 mothers' first schedule). Mothers who had already expressed some resentment in response to their situation, were significantly more likely to exhibit some negative attitudes to their C.F. children in these descriptions ($X^2 = 13.1$, d.f. = 1, $P < 0.001$).

Throughout this analysis, the coping ability of the mothers has been cited as a salient concept. It is useful now to summarise some of the points made thus far which can be subsumed under this heading. (Table III.D.(xiii)). For the purposes of this analysis, coping was conceived of as a tripartite index, in which the realistic appreciation of the disease and the adequate care of the child; the ability to fulfil other responsibilies and the ability to tolerate the situation without disruptive anxiety and depression were given equal weight. However it was not imagined that such an index, derived in a fairly arbitrary way, could provide a sensitive indicator of gradations in coping ability. The 18-point scale which emerged, was in fact, effectively reduced to two points, by designating scores 0-8 as evidence of poorer coping ability and scores 9, as better coping ability.

Table III.D.(xiii) To Derive an Index of Mothers' Ability to cope with C.F. (1 point awarded for each positive response - index values 00-18)

1. Realistic appreciation of the disease and adequate care of the child
   (a) Mother shows good understanding of the diagnosis (I,Q.103-107)
   (b) Mother does not doubt the diagnosis (II,Q.19)
   (c) Mother brings child regularly for treatment (from Hospital Records)
   (d) Mother spends an appropriate length of time in treatment (I,197)
   (e) Mother does not experiment with treatment prescribed (II,Q.25)
   (f) Mother has confidence in her ability to care for the child (I,199)

2. Ability to continue to fulfil other responsibilities
   (a) Mother does not neglect her other healthy children because of C.F. (I,Q.278)
   (b) Mother can still meet her other family responsibilities (II,Q.21)
   (c) Mother continues to plan for the future (I,Q.402)
   (d) Mother is not socially isolated because of C.F. (II,Q.27)
   (e) Mother does not have marital difficulties because of C.F.(or has had but has overcome them)(II,Q.42)

(f) Mother is not personally frustrated in her life by C.F. (II,Q.40)

3. Ability to tolerate the situation without disruptive anxiety or depression.

(a) Mother's health has not been disturbed by her response to her child's condition (II,Q1-5)

(b) Any disturbances which may have occurred have not been severe enough to warrant medical treatment (II,Q.1-5)

(c) Mother feels she can bear the burden of cystic fibrosis (II,Q.20)

(d) Mother is not always discouraged by C.F. (II,Q.22)

(e) Mother can maintain optimism for the child (II,Q.23)

(f) Mother's anxiety level is not unduly high (above average on Taylor's Scale i.e. 15)

Mothers' scores on this scale as a whole ranged from 1 to 16 and showed a mean of 11.5 and a standard deviation of 3.4.

Although the scale was somewhat arbitrarily derived the intercorrelations of its three major constituent parts are of considerable interest to this study. There was a significant correlation between the two practical aspects of coping behaviour, i.e. between ability to care adequately for the sick child and to fulfil other responsibilities (Pearson's $r = 0.29$, $N = 49$, $P < 0.02$). There was no significant correlation between the mother's ability to cope with her C.F. child and her ability to

tolerate the situation personally (Pearson's r = 0.10, N = 49, $P \triangleq 0.25$) but there was a high degree of correlation between the ability to fulfil other responsibilities and the mother's own ability to cope with the stress of her situation (Pearson's r = 0.59, N = 49, $P < 0.001$). Thus, a mother who is greatly disturbed by having a child with cystic fibrosis is likely, nevertheless, to be able to care adequately for that child; it is in the conduct of their other responsibilities that these highly anxious mothers may be found wanting. There is a tendency however, cutting across these correlations, for mothers who can cope adequately with their sick child, also to be able to cope with their other responsibilities.

Finally, in this chapter, since the influence of cystic fibrosis seemed to reach the most deeply personal aspects of the lives of the parents of these children, it was felt worthwhile to consider the role of religious beliefs in parents' response to their situation. Although religious faith was a very significant factor in helping a minority of parents to come to terms with their child's disease, almost an equal number of parents reported that, on the contrary, they felt their faith had been shaken by their experiences. Table III.D.(xiv).

Table III.D.(xiv). Religious Attitudes of C.F. Parents

Table III.D.(xiv). Religious Attitudes of C.F. Parents.

	Mothers	Fathers
Religion has helped parent to come to terms with C.F.	22%	23%
Religious beliefs questioned because of C.F.	29%	11%
	Total N = 49 mothers	Total N = 47 fathers

Although the number of Roman Catholic families in the sample was small, it was of interest that only one of the eight Roman Catholic parents reported having had doubts about his religious beliefs as a consequence of cystic fibrosis although only half of them felt that their faith had positively helped them to face their problems.

Having described the background in terms of medical history, family variables and parental reactions, the way is now clear for us to focus directly on the problems occasioned by cystic fibrosis for the growing child.

E. The Influence of Cystic Fibrosis on the Development of Affected Children

To some extent, the title of this chapter is a misnomer, for the scope of the study reported here did not extent to a longitudinal study of C.F. children. The heterogeneous nature of its sample also ruled out the possibility of a carefully controlled cross-sectional design, so that we cannot truly claim to have followed the developmental course of the influence of the disease on children. However, the systematic gathering of information, in a cumulative way, about the development of all the children in the sample, from birth to the time of the enquiry, allows us to present, at least, a preliminary report of the influence of cystic fibrosis on a number of aspects of child development.

(1) Infant development (children < 5:0 years)

(2) C.F. Children and their illness

(3) The Education of Older Children ($\geq$ 5:0 years)

(4) The Social and Emotional Development of Older Children ($\geq$ 5:0 years)

There were insufficient numbers of adolescents in the sample to afford any systematic examination of the special problems of their age-group in relation to cystic fibrosis. Nevertheless, these problems do seem to evolve from difficulties experienced in earlier years and to this extent the chapter may be usefully summarised and concluded by a brief note on the experiences of the young people who assisted in this study.

1. Infant Development (Children < 5:0 years)

Preschool development was assessed solely from mothers' reports. In the case of the absentee mother, the grandmother, who had cared for the child before and since his mother's departure, and the father, who was unemployed and at home all day, were able to supply most of the information required. Most of these findings then, refer to the preschool development of all the 58 children in the sample. It should be recalled, though, that for 37 of these children, now over the age of five years, this information is retrospective, while the remaining 21 children have not yet passed through the whole of this period of development. In some instances, only those findings which refer to the children who were infants at the time of the enquiry, are appropriate.

Before the developmental history of each child was investigated, mothers were asked a few questions about their own ante-natal history, in order to assess the nature and degree of stress to which they had been subject, during that pregnancy. This was of interest, in relation to a subsidiary hypothesis, that the stress experienced by mothers during pregnancy would be a significant factor in the incidence of feeding difficulties and vomiting among their babies. A nine-point scale evolved from mothers' responses to the questions indicated in Table III.E.(i).

Table III.E.(i) Sources of Maternal Stress in Pregnancy

Table III.E.(i) Sources of Maternal Stress in Pregnancy

I.287 Were you at all emotionally upset during your pregnancy?

288 How did your husband feel about the pregnancy - was he pleased or displeased?

289 Had you sustained any miscarriages or lost any babies before this pregnancy?

290 Did you know about C.F. at this stage?

291 Did you suspect that N might be a C.F. child?

292 Did you lose anyone close to you during your pregnancy?

294 Did you move house during this pregnancy?

296 Was your husband in full employment during this pregnancy?

These 8 questions were supplemented by the consideration of whether or not the child had been conceived out of wedlock. Illegitimacy and marriages of necessity were apparent from mothers' comments and from the dates of marriage and birth recorded at the beginning of the interview (I.Q.3 and 18)

Maternal stress, as experienced during these 58 pregnancies, ranged from 0-6 on the scale and showed a mean index value of 1.7, with a standard deviation of 1.2. No significant relationship was found between pregnancy stress, assessed in this way, and the infants' early feeding difficulties ($\chi^2 = 1.59$, d.f. = 2, $P \triangleq 0.50$).

At birth, the C.F. children appeared physically normal although, as we have already indicated, three of them were

suffering from a bowel blockage which required surgical intervention soon afterwards.

On the whole, the children were found to pass the milestones of motor and verbal development at appropriate ages. Although infants who were very ill, and/or in hospital for relatively long periods at this time, inevitably fell behind in the acquisition of new skills, these delays were not specific to their condition. There was no evidence, then, that cystic fibrosis per se, had influenced the children's early development in these respects.

In describing the influence of cystic fibrosis on the development of the preschool child, the intention was not to reiterate an account of the symptoms presented by the undiagnosed child, so that the remainder of this section will, as far as is possible, refer only to those problems which persisted post-diagnostically.

There was some evidence that the customary problems of childhood were somewhat exaggerated by cystic fibrosis, for example in feeding, toilet-training and general behaviour. Illingworth (1972) has cited some comparable figures, referring to the incidence of these problems in his experience of normal children, ranging in age from 2-5 years. These are quoted in Table III.E.(ii) to provide a standard by which the findings of this study may be evaluated.

Table III.E.(ii). The Incidence of Childhood Problems among C.F. Infants.

Table III.E.(ii). The Incidence of Childhood Problems among C.F. Infants.

	C.F. Infants	Normal Infants*
Feeding problems	41%	10 - 20%
Toilet-training problems	63%	10 - 20%
Behaviour problems	55%	20 - 30%
	N = 58	*from Illingworth

Contrary to the classical picture of the disease, the children posed feeding problems because of their poor, rather than voracious appetites. They also presented toilet training problems, particularly with reference to soiling, and behaviour problems of attention-seeking, clinging to mother and inordinately demanding behaviour. Problems in this aspect of the study were not defined by objective criteria but were, rather, scored when a mother considered this to be an area of difficulty in her rearing of her C.F. child. Although these were problems which did persist post-diagnostically, they may still be partly understood with reference to the child's symptoms. This is particularly true of the toilet-training problems where very real difficulties such as bowel prolapse, may occur.

The behaviour problems seemed to extend to bed-time, and when combined with the children's reported timidity, they presented as problems connected with getting the child to sleep or with sleep itself. Almost half of the children (47%) had presented problems of this nature during infancy. Fears of being left alone in the dark were common. Mothers reported fairly frequent

nightmares and spells when their children would awake crying during the night. As a result, 19 of the total number of children in the sample were regularly taken into bed beside their parents during infancy and the same pattern was borne out by the 7 of the 21 children who were infants at the time of the enquiry. These findings are harder to evaluate, since the Newsons report that for one child in five, among normal four year olds, waking in the night is not at all uncommon, and that the proportion of mothers who take their children into bed with them, when this happens, is indeed high.

Significant factors in the nature and extent of the difficulties presented by these infants can only meaningfully be assessed in relation to those 21 children who were under five years of age at the time of the study. Since the sample was so unabalanced with respect to the sex of the children, being composed of 18 boys and only three girls, it was not possible to assess the significance of sex differences in the incidence of infant difficulties reported.

The clinical condition of the infant is undoubtedly a significant factor in the number and severity of the difficulties he presents. Cystic fibrosis children are not easy to rear and those who are most severely affected are more likely to present several problems of the nature of those discussed above (Table III.E.(iii).

Table III.E.(iii). Infant Difficulties and the Severity of the Child's Condition.

Table III.E.(iii). Infant Difficulties and the Severity of the Child's Condition.

	A	B	C
Child presents a single problem or no problems at all	4	1	0
Child presents several problems	8	6	2
	Total N = 21 infants (aged 1 - 5 years)		

At the same time it is interesting to note that the mothers who were highly anxious were twice as likely to report multiple problems, as were the mothers who were less anxious, and that the mothers who described marital difficulties were also more likely to encounter several difficulties during their child's infancy. However, these trends, apparent in this small sample, will require cautious interpretation in the discussion of the findings. (Section IV).

In an attempt to assess mothers' attitudes to their difficult infants, all of the mothers in the sample were invited to describe their perceptions of their children at this age. Mothers who described their child only in negative terms were said to be displaying a negative attitude to the child. Positive and mixed feelings were grouped together in a single category. All the attitudes expressed by the mothers were assessed at face value. On this basis, mothers who were resentful of the influence of cystic fibrosis in their lives were significantly more likely to describe their children, at the infant stage, in

solely negative terms (X^2 = 13.1, d.f. = 1, $P < 0.001$).

The personal and social development of the infants in the sample was described with the aid of the Vineland Social Maturity Scale. The infants showed a fairly normal distribution of social quotients, ranging from 75 to 138 with a mean of 104 and a standard deviation of 10.3. This obscured a trend for the children to fall behind the norms in the category of the development of independence or self-help, because, on the mothers' own admission, their mothers did more for them than was really necessary. One third of the children also obtained lower scores than those expected of children of their age, in respect of the category of socialisation. Although the American norms for this scale may not be entirely appropriate for use in British samples, this was an area in which the mothers had already recorded some difficulty with their C.F. children. These children then, did tend not to play with other children, but in this context it is interesting to recall that one in four of the mothers of these had already said that they expected less of their C.F. children in terms of their relationships with other children.

Clearly we must acknowledge that the physical condition of the C.F. infant may create a number of problems during his pre-school years. Although the nature of the sample restricts the generalisations that can be made from this section of the findings, it will be important to include in the discussion of these findings some consideration of the role of maternal anxiety in these early difficulties. It is important to reach the roots of these infantile problems and to search for means of alleviating

them, for the incidence of such difficulties has already been shown to be highly influential in parents' subsequent attitudes to the upbringing of their C.F. children. (Mothers, $X^2 = 8.98$, d.f. = 1, $P < 0.01$).

2. C.F. Children and their Illness

During the first interview, mothers had been asked about the nature and troublesomeness of their children's symptoms. (I,Q.244-249) Their responses, which are recorded in Table III.E.(iv), were considered to be important for our understanding of the influence of the disease on the children.

Table III.E.(iv) The Symptoms Exhibited by C.F. Children.

	Symptom present but not troublesome	Symptom is/ has been troublesome
Nasal discharge	17%	21%
Bad cough	14%	38%
Foul-smelling stools	45%	33%
Large appetite	16%	9%
Abdominal pain	16%	43%
Bowel prolapse	3%	34%
	Total N = 58 children	

Although an attempt was made to gather some information in a systematic way, about the children's own awareness of their

symptoms there was a considerable degree of variance in the responsiveness of the children in this matter and the findings that emerged are better included in the discussion of the results, by way of amplification of parents observations.

Before considering the reactions of these children to their illness it was important to ascertain how much factual information the children were likely to have about cystic fibrosis. Mothers' assessments about the information which they thought their children had about the disease are listed in Table III.E.(v). In the matter of factual information it was slightly easier to score the children's own responses and alongside these assessments are given the proportions of children who indicated by their conversations with the investigator that they did indeed possess the information in question.

Table III.E.(v). Information C.F. Children have about Cystic Fibrosis.

	Mothers	Investigator
Child knows the name cystic fibrosis?	29%	33%
Knows he was born with it?	31%	not asked
Knows it was inherited?	10%	not asked
Knows how it affects his body?	45%	49%
Knows he will always have it?	38%	57%
Knows anything of the prognosis?	10%	22%
	from reports of 58 children	from conversations with 37 children

If the child had some age-appropriate knowledge of the effect of the disease on his body and was aware that he would always have this problem and always require treatment for it, then his understanding of cystic fibrosis was scored as 'adequate'. If he knew more than this, it was scored 'good' and if he knew less it was scored 'poor'. Children who knew nothing at all, and those who were too young, in their mothers' estimation, for these questions to be relevant, were scored in the category 'nil'. Thus the distribution of knowledge on these two estimates, as before, may be seen in Table III.E.(vi).

Table III.E.(vi). Assessments of Children's Knowledge of Cystic Fibrosis.

	Mothers	Investigator
Good	28%	28%
Adequate	17%	16%
Poor	16%	28%
Nil/Too Young	39%	28%
	referring to 58 children	referring to 37 children

All children under the age of four years were considered by their mothers to be too young to have any appreciation of their condition. Between the ages of 4 and 10 years there is no significant age relationship in the knowledge which the children

possess but among the children who were 10 years old or older, only one child in twelve had a less than adequate knowledge of the illness. Factors other than the age of the child were also found to be influential. Among those over the age of four years, the children who had siblings also affected by C.F. tended to know more about the disease than children whose siblings were all healthy, but the trend did not reach statistical significance ($X^2 = 3.58$, d.f. = 1, $P < 0.10$).

Surprisingly, the relationship between the children's knowledge and their level of communication with their parents did not reach statistical significance (Mothers, $X^2 = 1.54$, d.f. = 1, $P \simeq 0.20$; Fathers, $X^2 = 0.44$, d.f. = 1, $P \simeq 0.50$). This could be a reflection of parents' later admission of the difficulties they experienced in answering their children's questions about cystic fibrosis. Only 51% of all the parents in the sample felt able to deal with such questions with ease and truthfulness.

Parents had suggested that the children, as they grew older, "picked up a lot", during clinic visits and hospital admissions. This assertion is hard to test but it would account for the absence of other significant relationships of factors with the children's knowledge of C.F.

The reactions of the children in the sample, to their illness and to its implications for them, may be appreciated from the observations of parents which were gathered with the aid of questions 234 to 242 on the mothers' first schedule and of questions 51 to 59 on the fathers' schedule. These observations are recorded in Table III.E.(vii), although fathers' descriptions

are missing for three of the children where these fathers were not available for interview.

Table III.E.(vii) Parents' Observations of C.F. Children's Reactions to their Illness

Child is:	Mothers	Fathers	Agreement between Parents
Alarmed by C.F.?	16%	5%	X^2 =17.5;d.f.=2,P<0.001
Pleased by special attention?	36%	33%	X^2 =29.1;d.f.=2,P<0.001
A hypochondriac?	36%	36%	X^2 =10.1;d.f.=2,P<0.01
Self-conscious?	40%	18%	X^2 =14.7;d.f.=2,P<0.001
Frightened because C.F. makes him different from others	4%	5%	X^2 =18.0;d.f.=2,P<0.001
Resentful because C.F. makes him different from others	26%	22%	X^2 =10.2;d.f.=2,P<0.01
Sad because C.F. makes him different from others	16%	10%	X^2 =23.6;d.f.=2,P<0.001
Worried about his size or shape?	28%	16%	X^2 =22.2;d.f.=2,P<0.001
Given to periods of increased worry about himself	12%	13%	X^2 =29.0;d.f.=2,P<0.001
Liable to show an exacerbation of symptoms if upset	19%	10%	X^2 =4.68;d.f.=2,P<0.10
	(refers to N = 58 children)		

From the number, the duration and the severity of the children's reactions, as observed by their parents, it was possible to assign a single global assessment to describe their reactions to cystic fibrosis. On the basis of mothers' comments 14 of the

children in the sample, i.e. 24%, were described as showing a marked or severe reactions to their illness. These children exhibited a number of reactions of some permanence in their response repertoire, to an extent which caused the mothers concern. 13 of the children, 22% of the sample, showed moderate or less severe reactions than those just described. Children with slight reactions tended to show reactions to their illness only sporadically, and then not in an extreme way. 12 such children were identified (21%). The remaining 33% of the sample showed no reactions at all.

A number of factors appeared to be significantly associated with the severity of the children's responses. Certainly to some extent, the children's reactions must be seen as reasonable responses to the situation in which they find themselves, for the degree to which their symptoms are troublesome ($X^2 = 8.97$, d.f. = 1, $P < 0.01$) and the severity of their condition as clinically assessed, ($X^2 = 8.37$, d.f. = 2, $P < 0.02$) showed highly significant relationships with their behaviour as described by their parents.

The child's knowledge of his condition seemed to be an important contributory factor, since the more the child knew of cystic fibrosis the more marked was likely to be his reaction ($X^2 = 5.49$, d.f. = 1, $P < 0.02$) and this was especially true of the single factor of whether or not the child had ever been exposed to the possibility of a fatal outcome to his disease ($X^2 = 9.86$, d.f. = 1, $P < 0.01$). Family communication certainly seemed to have some influence in the matter of the child's reactions for in families where communication, as we have

described it, was good, the children were much less likely to react severely to their condition, but on analysis the trend did not reach statistical significance ($X^2 = 3.89$, d.f. = 2, $P \triangleq 0.20$).

Although the nature of the observations made by parents can best be amplified in the discussion by cross-reference to findings from conversations with the children themselves, it is interesting to give pause for a moment to consider the developmental pattern of reaction which is suggested by these findings, although there is no significant age difference in the severity of the children's responses reported ($X^2 = 4.54$, d.f. = 2, $P \triangleq 0.10$).

The response of the young child seems most likely to be one of pleasure at the special attention that is his, because of his health and it seems significant that this reaction is strongly associated with the observation that when the child is upset by something his C.F. symptoms tend to be exacerbated ($X^2 = 9.91$, d.f. = 1, $P < 0.01$).

After the child goes to school, clusters of response tendencies begin to emerge. Anxiety about his physique may certainly make the child self-conscious ($X^2 = 14.5$, d.f. =1, $P < 0.001$) and the child who feels anxious about his size or shape is more likely to become alarmed about C.F. ($X^2 = 10.6$, d.f. = 1, $P < 0.01$). As he becomes alarmed about the disease the child is more likely to undergo periods of worry about himself ($X^2 = 24.1$, d.f. = 1, $P < 0.001$). Certainly, as the child grows older reports of his resentment at being different from others become more likely, although the more anxious

children respond to being different by fear or sadness rather than resentment . (Anxiety about self x fear, $X^2 = 7.73$, d.f. = 1, $P < 0.01$; anxiety about self x sadness, $X^2 = 14.4$, d.f. = 1, $P < 0.001$; anxiety about self x resentment, $X^2 = 0.58$, d.f. = 1, $P \simeq 0.50$).

The consideration of the children's reactions to their illness may be of importance as an intervening variable in the interpretation of the influence of cystic fibrosis on their development. We turn first, to the intellectual development of the school children in the sample to consider the influence of the disease on their intellectual ability and on their scholastic performance.

3. The Education of Older Children ($\geq$ 5:0 years)

Parents of chronically ill children commonly express anxiety about the effects of the illness on the child's mental capabilities and on his education so that some attention was given to these matters in the course of this study.

Thirty three children were tested individually in their homes on the Wechsler Intelligence Scale for Children. They attained scores which showed an approximately normal distribution, ranging from I.Q.'s of 74 to 138, and having a mean of 105 and a standard deviation of 14. The boys in the sample appeared rather more intelligent than the girls, showing a mean I.Q. of 107 as against the girls' mean I.Q. of 102.

It had been suggested in the literature that C.F. children appear more intelligent than they really are, by being

highly articulate, and that this increase in verbalisation reflects a raised level of anxiety. If the children's verbal skills were particularly well developed for their years it was expected that this would be reflected in improved scores on the verbal scale of the W.I.S.C. The children's verbal and performance scores on this scale are compared in Table III.E.(viii).

Table III.E.(viii). The Verbal vs. Performance Scores of C.F. Children (from the W.I.S.C.)

	Girls	Boys	Total (boys and girls)
$\geqslant$10 points difference, V $\gg$ P	20%	11%	
> 2, < 10 points difference, V > P	20%	21%	37%
V = P $\pm$ 2 points	20%	6%	12%
> 2, < 10 points difference, P > V	7%	6%	51%
$\geqslant$10 points difference, P $\gg$ V	33%	56%	
	= 15	= 18	N = 33 children

Although there was a tendency for some children, particularly girls, to obtain scores which exemplified this trend, the results as a whole show that in this sample there was a strong counter-trend for the children to show more highly developed performance skills.

On balance, then, the hypothesis was not confirmed by the findings from this sample.

All of the children attended normal day schools at the time of the enquiry, although one boy had begun his education in a special school for handicapped children and two children had had spells of home teaching during prolonged absence from school. Most of the mothers were happy with this arrangement although 25% of them were anxious about the risk thereby imposed, of exposing the child to infections. Six of these eight mothers admitted that this anxiety encouraged them to keep their children away from school and three definitely felt they would prefer to have some educational provision made for the child for these reasons. Home teaching seemed to them to be indicated.

On the whole though the school attendance of the children was fairly good. Although their attendances tended to be poorer during the winter months, as many as 70% of these children were described as having fairly good records of school attendance.

To assess school achievement, tests of arithmetic, reading and spelling were given, again in the child's home, and scored against age related norms. Retardation was expressed in months and the mean retardation scores for the whole sample, in these three basic subjects are given in Table III.E.(ix).

Table III.E.(ix). Mean Retardation Scores of C.F. School Children in Basic School Subjects (expressed in months).

Table III.E.(ix). Mean Retardation Scores of C.F. School Children in Basic School Subjects (expressed in months.)

	Mean Retardation	Standard Deviation
Arithmetic	13.1 months	11.3
Reading	9.3 months	11.1
Spelling	11.4 months	13.0
		Mean Retardation over 3 subjects : 11.3 months (S.D. = 11.8)
	N = 33 C.F. School Children	

Surprisingly enough only 20% of the children were rated as being poor at arithmetic by their class teachers, the remainder of the sample being described as average or better. 26% were described as being poor at reading but there was no significant difference between the ratings given to these children and their controls, matched for ability, by the teachers. (Arithmetic, $X^2 = 0.08$, d.f. 1, $P \simeq 0.80$; Reading, $X^2 = 0.80$, d.f. = 1, $P \simeq 0.30$).

To allow then for possible variations between the standards of the children's schools and for possible variations of the relevance of the tests to the children's curricula, a child was then said to be underachieving, for the purposes of this study, only when his test score fell below that expected of a child of his age and intelligence, by twelve months or more. It was also allowed that any child may have a weak subjects. Thus, only those children who showed such gross evidence of under-achievement in more than one of these basic subjects, were included in Table III.E.(x).

Table III.E.(x). Indications of Intellectual Underfunctioning among C.F. School Children.

	Girls	Boys
Is underachieving, by 12 months, in more than 1 basic subject	28%	44%
Average retardation shown over scores in all 3 subjects	9 months	13 months
	= 15 girls	= 18 boys
	Total N = 33 children	

Even with these generous concessions there was still some evidence of poor school work in basic subjects, among C.F. children and the problem seemed to be more common and more severe among the boys in the sample. A number of possible reasons for this academic underachievement were explored.

Where schooling has been interrupted by frequent or long hospitalisation we should expect some retardation in school work, but this correlation only reached statistical significance in the case of arithmetic scores. (Duration of hospital experience x retardation in arithmetic, Pearson's r = 0.40, sig. level = 0.01). However the differences in school performance between children graded A, B and C on the clinical scale were not significant,

suggesting that other factors are also in operation (Arithmetic, $X^2 = 0.49$, d.f. = 2, $P \simeq 0.80$; Reading, $X^2 = 0.78$, d.f. = 2, $P \simeq 0.70$; Spelling, $X^2 = 0.79$, d.f. = 2, $P \simeq 0.70$).

Deafness as an additional complication of cystic fibrosis has been recorded, but few of the children in this sample showed any hearing loss at all, and those who did were only very mildly deaf so that in no case was the hearing loss of sufficient severity to account for poor school performance.

We have already observed that poor school work seemed to be more characteristic of boys than of girls. However, no statistically significant differences were found between the performance scores of boys and girls although there was a trend, most pronounced in reading, for girls to be less likely to be retarded in their performance than were the boys ($X^2 = 1.89$, d.f. = 1, $P \simeq 0.20$).

Paradoxically there was a highly significant positive correlation between high intelligence quotients and high retardation scores, suggesting that the more intelligent the child, the more likely it seemed to be that he would not fulfil his potential in academic attainment tests. Table III.E.(xi).

Table III.E.(xi). Correlations between I.Q. and Degree of Retardation in School Subjects among C.F. School Children

	Pearson's r	Significance Level
I.Q. x Retardation in Arithmetic	0.41	0.006
I.Q. x Retardation in Reading	0.40	0.007
I.Q. x Retardation in Spelling	0.44	0.003

Emotional upset is recognised as being a causative factor in poor school work and we will discuss the findings concerning the emotional development of these children, presently. For the moment it is interesting to note that the children who were aware of the possible outcome of their illness were rather more likely to obtain poor scores in tests of school work, particularly in tests of verbal skills. (Reading, $X^2 = 2.67$, d.f. = 1, $P \simeq 0.10$; Spelling, $X^2 = 5.97$, d.f. = 1, $P < 0.02$).

It was interesting then, to consider the role of parental attitudes in the children's school performance. It will be recalled that some parents did report a lowering of their expectations of their C.F. children in terms of school achievement. For simplicity these views were summarised in an index of parental attitudes, the derivation of which is figured in Table III.E.(xii). With a point added to the total for each positive response the index took values from 0 to 6, representing increasingly permissive parental attitudes towards the schooling of their C.F. children.

Table III.E.(xii) To Derive an Index of Parents' Attitudes to the Schooling of their C.F. Children.

Table III.E.(xii) To Derive an Index of Parents' Attitudes to the Schooling of their C.F. Children.

*I.255.	Mother expects less of the child in terms of school achievement?
I.255.b.	Mother feels protective of the child in this sense?
I.391.a.	Mother sometimes keeps the child at home in response to her own apprehension?
I.392.	Mother would like other educational provision to be made for him?
III. 64	Father expects less of the child in terms of school achievement?
III. 64.b.	Father feels protective of the child in this sense?

Index takes values 0-6

*References refer to question numbers on the interview schedules in the Appendix

Interestingly the correlation between parental permissiveness and poor school work was also highly significant (Pearson's r = 0.41, sig. level = 0.006).

Our sample was not large enough to permit meaningful partial correlations but the findings are sufficient to suggest that there are several factors which can mediate the influence of cystic fibrosis on the school performance of affected children.

Thus far we have neglected to consider the practicalities which may present obstacles for the C.F. child in going to school.

Questions of a practical nature were uppermost in the minds of many mothers and it was clear from the interviews that the anticipatory level of anxiety about these problems was particularly high among the mothers of pre-school children. Once the children started school these anxieties were somewhat alleviated but some practical problems did arise which should not be overlooked by this study. Table III.E.(xiii).

Table III.E.(xiii) The Incidence of School Problems among C.F. Children

	Boys	Girls	Total
Initially had problems settling in	32%	28%	30%
(% directly related to C.F.)	(50%)	(-)	8%
Has had school problems since	37%	17%	27%
(% directly related to C.F.)	(43%)	(33%)	11%
	N = 19 boys	N = 18 girls	N = 37 children

From the mothers' reports then the majority of the children in this sample had not presented serious school problems. Almost a third of the sample had been very unhappy about leaving their mothers to go to school but mothers were prepared to accept this as a normal response and only 8% of the children in the sample had had C.F. related problems on starting school e.g. toilet difficulties.

Toilet related difficulties persisted in the older age group and 11% of the mothers reported such problems while a further 16% reported problems of children playing truant.

On the whole, mothers reported having had good cooperation from the children's school teachers. 14 of the mothers had felt that the teachers had been unduly worried about having a C.F. child in the class but in only 6 cases did mothers feel that this had lead to the child being given extra attention, although a further 4 mothers had made a visit to the school expressly to ask that this should not happen. Although 9 mothers felt that at least one of their child's teachers had expected less of the child because of his illness this showed a strong relationship to the severity of the child's condition ($X^2 = 6.45$, d.f. = 2, $P < 0.05$) and these mothers were quite grateful for it. Happily, it was rare for mothers to find teachers who were unduly troubled by the child's symptoms, 4 out of 37, or who 'picked on' the child because of them, 1 in 37.

The majority of the children, 78% of them, seemed enthusiastic about school in conversation with the investigator and none of them volunteered information about the problems raised for them by cystic fibrosis. However, when asked specifically 24% of the children admitted considerable embarrassment about toilet problems at school and the nature of the topic and their embarrassment suggested that this was an underestimate of the true incidence of problems. Coughs and tummy pains providing nuisance value at school were more readily mentioned by a further 31% of the children, while 13 of the 21 children who had lunch at school were acutely embarrassed by having to take their

medicine before the meal in front of their peers. They resolved their difficulties by seeking solitude to take them or by disposing of the pills into the waste paper bin.

Although these findings concerning the practical problems for the C.F. child at school are not amenable to neat numerical analysis they are nevertheless vitally important to our understanding of the children as they are described in the next part of this chapter.

4. The Social and Emotional Development of Older Children.

A hint of disturbance in the social development had already been caught in the low 'Socialisation' scores obtained by the pre-school C.F. children on the Vineland Social Maturity Scale and this trend was continued in the descriptions given by mothers of their older C.F. children. Table III.E.(xiv)

Table III.E.(xiv). The Social Activities of C.F. Children.

	Boys	Girls	Total
Prefers solitary play	68%	44%	57%
Always wants to play in own home	53%	44%	49%
Cannot stand up for himself	42%	33%	38%
Takes an active part in sport	47%	50%	49%
	N = 19 boys	N = 18 girls	N = 37 children

Thus, at an age when most children are becoming increasingly sociable and enjoying increasing freedom of movement outside their own homes, C.F. children are often still to be found playing alone and at home and this was often particularly true of the boys in this sample. This social isolation was to some extent, broken for some of the children by their participation in some kind of sporting activity. Participation in sport was actively encouraged by the children's clinic doctors.

The children's social development was summarised, for the purposes of statistical analysis, into a single index, on the basis of mothers' reports. Table III.E.(xv). The interpretations of the findings of the analysis will again be tempered, in the discussion, by reference to the children's own observations.

Table III.E.(xv). To Derive an Index of Social Development

Child's communication with others outside the family is generally poor?
Child plays alone?
Child plays at home?
Child is unable to stand up for himself against his peers?
Child has not made friends at school?
Summed positive responses give index value 0 - 5

The school children in the sample obtained values of this index which ranged from 1 to 5 and showed a mean of 1.8 and a standard deviation of 1.5 No significant sex differences were found in the index values assigned to the children (X^2 = 0.60, d.f. = 1, P $\simeq$ 0.50) but the most severely affected children did seem most likely to show poor social development (X^2 = 7.49, d.f. = 2, P < 0.05).

Again the most significant factor in the children's social development seemed to be parental expectations. The relevant parental attitudes were again drawn together and simplified into an index as before. Each positive response added one point to the total score which then ranged from 0 to 6 expressing decreasing parental pressure towards the child's normal social development. Table III.E.(xvi).

Table III.E.(xvi) To Derive an Index of Parental Attitudes to the Social Development of C.F. Children

*I.256./III.65.	Mother/Father expects less of the child in his relationships with other children)
I.257/III.67	Mother/Father permits the child less independence in joining activities outside his own home
I.258/III.68	Mother/Father is more cautious about allowing the child to join in physical activities.

* refers to questions in Appendix. Index takes value 0-6

Again the more anxious parents are about their C.F. child's activities the more likely ti becomes that his social development, as assessed here, will be abnormal. (Pearson's r = 0.37, sig. level = 0.01).

Away from his parents' protectiveness, we can assess the child's social adjustment from his school behaviour, by means of Stott's Bristol Guide for 'the Child in School'. Teachers' descriptions of the children at school were available for 35 of the C.F. children in the sample, and for 35 matched normal children each selected from the same class as the child inq uestion, to provide a control for local variations in standards of behaviour expected.

The cut-off scores suggested by Stott's norms were used in Table III.E.(xvii) to give a first impression of the distribution of scores in scales for under-reactive and over-reactive behaviour (Stott's unract and ovract). Thus:

	Cut-off scores for unract	Cut-off scores for ovract
0 = stability and near stability	0-2	0-3
1 = mild under/over-reaction	3-5	4-7
2 = appreciable under/over-reaction	6-8	8-11
3 = maladjusted under/over-reaction	9-14	12-24
4 = severe maladjusted under/over-reaction	15+	25+

Table III.E.(xvii) Distribution of Scores of C.F. Children and their Controls on Stott's Unract-Ovract Scales

Table III.E.(xvii) Distribution of Scores of C.F. Children and their Controls on Stott's Unract-Ovract Scales

Unract	0	1	2	3	4
C.F. group	23	5		1	6
Control group	29	2	2	1	1
Ovract					
C.F. group	20	5	5	3	2
Control group	27	1	1	3	3
	N (C.F. Group) = 35; N (Control Group) = 35				

On the basis of this first analysis then, there was some evidence that the C.F. children at school were more likely to show under-reactive and over-reactive. 32% of the C.F. group were described as showing some degree of under-reactive behaviour as sompared with only 16% of the control group, while in terms of over-reactive behaviour, 45% had obtained some score for behaviour disturbance while only 22% of their controls had been so described. The mean scores obtained by the groups on these scales are shown, separated by sex in Table III.E.(xviii). The mean scores quoted by Stott are also given.

Table III.E.(xviii) Mean Scores obtained by C.F. Boys and Girls and their matched controls on Stott's Scales of Unract and Ovract.

Table III.E.(xviii) Mean Scores obtained by C.F. Boys and Girls and their matched controls on Stott's Scales of Unract and Ovract.

	Unract	Ovract
C.F. Boys (N=18)	M=8.61, S.D.=15.2	M=8.27; S.D.=12.4
Control Boys (N=18)	M=0.55; S.D.=2.11	M=8.11; S.D.=12.4
Normal Boys (Stott)	M=3.05; S.D.=3.66	M=5.11; S.D.=6.60
C.F.Girls (N=17)	M=3.17; S.D.=6.89	M=3.64; S.D.=5.45
Control Girls (N=17)	M=1.94; S.D.=3.95	M=1.35; S.D.=2.34
Normal Girls (Stott)	M=2.84; S.D.=3.83	M=2.71; S.D.=4.80

The most striking finding to emerge from teachers' assessments of these children was, then, the tendency for C.F. boys to be more likely to show under-reactive behaviour, and to do so to a greater degree, than either their controls or the C.F. girls.

The number of children obtaining these scores was small in statistical terms, so that the discussion of the precursors of good and poor adjustment has to depend largely on insights from parents, and these will be included in the interpretation of the results, given in the next Section.

Although parents accounts of their children's reactions to their illness have already given some indication of the emotional difficulties of these children, the study encompassed three further sources of information on this matter.

29 of the children in the sample were able to complete the Children's Form of the Manifest Anxiety Scale. They attained scores which ranged from 6 to 34 and showed a mean of

18.1 and a standard deviation of 7.07. Separate means for boys and girls are shown in Table III.E.(xix) with the means quoted by Castaneda et al (1956) for normal children.

Table III.E.(xix) Mean Scores of C.F. and Normal Children on the Children's Manifest Anxiety Scale

		Boys	Girls
Cystic Fibrosis Children (N = 29)	Mean:	18.4	17.8
	S.D.:	7.03	8.02
		(N = 17)	(N = 12)
Normal Children (from Castaneda et al)	Mean:	18.45	15.9

The children's anxiety scores did not show any significant relationship to the severity of their clinical condition nor to their age (clinical condition, $X^2 = 0.82$, d.f. = 2, $P \doteq 0.70$; age, $X^2 = 0.07$, d.f. = 1, $P \doteq 0.80$). The relationship between the anxiety of mothers and that of their children did not reach statistical significance ($X^2 = 1.43$, d.f. = 1, $P \doteq 0.20$) although level of communication achieved between mother and child did seem to be a significant factor ($X^2 = 3.96$, d.f. = 1, $P < 0.05$).

Certainly some of the mothers were concerned about their children's emotional development. More than half of the mothers (54%) of these older children remarked that their children had

never grown out of their preschool timidity and 8 of these 19 older boys and 5 of the 18 older girls were described as moderately or very fearful at the time of the enquiry. Although the mothers of approximately 75% of these children described their children as generally happy there was a tendency for mothers of boys to describe their sons as 'moody' and their daughters as 'fretful' or 'whining' (First schedule, Q.33 was frequently responded to with reference to the child's temperament.)

16% of the children interviewed did themselves admit that they felt different from other children but even after wide-ranging discussions with these 37 children, about their wishes and dreams, fears and feelings, the investigator was unable to present evidence of the preoccupation with death, which the literature found so pervasive among these children. By the same token, no evidence was found of the distortions of the body image in the children's drawings which were interpreted in the literature as being indicative of emotional difficulties.

Although the reports in the literature seem to overstate the problems as experienced by the children in this study, these findings do nevertheless suggest that cystic fibrosis is not without influence in the development of these children. This conclusion is endorsed by the situation in which the older children in the sample were found.

The two young people who had left school were both unemployed at the time of the enquiry and the seven other older children ($\geq$ 11:0years) were realistically concerned about the limits imposed on their employability by cystic fibrosis.

Four boys in this older age group were very isolated from peer group activities, although three of the four girls in this group had successfully avoided social isolation by taking an active part in some sport, and their social life was more normal for girls of their age. Four of this group of nine looked strikingly young for their years and all of them had had spells of extreme sensitivity about their physical appearance in relation to cystic fibrosis. In the cases of two of the girls and two of the boys, mothers had particularly expressed anxiety about their child's reaction to the disease although in two of these cases the situation had improved by the time of the enquiry. These young people were themselves on the whole, very frank in their discussions with the investigator of the frustrations, the anxieties and the upsets of having cystic fibrosis.

It is hoped that the use of their observations in the interpretation of the other findings reported in this chapter, will give a better insight into the nature of the influence of cystic fibrosis over the development of these children and into the ways in which their problems may be alleviated.

Before these findings can be interpreted and discussed we have, finally, to consider the evidence which will allow us to assess the nature and the extent of the implications of the family situation which we have just described, for the other healthy children in the family.

F. The Healthy Siblings.

If the families of handicapped children have, until relatively recently, been rather neglected by research workers, then the healthy brothers and sisters of these children must surely represent the most neglected members of these families. The thorough investigation of the situation of these children would provide a complete research project in itself, if adequate attention were to be given to the important factors of the ordinal position, age and sex of the child relative to his ill sibling. Clearly such a comprehensive analysis was outwith the scope of this study, so that the attention given to the healthy siblings of the children in this sample is but cursory. Nevertheless, their existence has at least been acknowledged and a brief review of the findings of this study may assist in the definition of an area worthy of closer attention.

Only children and their families were, clearly, excluded from this and the case of families having two C.F. children will be described, with the interpretation of the results presented here, in the next Section. This chapter, then,is concerned with the 39 families in the sample who had one C.F. child and one or more other healthy children. The distributions of the sexes and of the ages of the children relative to their C.F. siblings, are given in Table III.F.(i).

Table III.F.(i) The Age and Sex Distribution of the Healthy Siblings in the Sample.

Total No. of Siblings	72
No. of brothers	44
No. of sisters	28
No. of Elder siblings	40
No. of Younger siblings	32

The existence of elder siblings already living away from home and younger siblings too young to convey their feelings unambiguously, meant that the numbers of children to whom these findings refer, are frequently less than these totals and are indicated as such.

Mothers were asked whether they thought that their other children had felt left out because of the extra attention the C.F. children required, particularly from their mothers. Although 62% of the mothers said they had explained to their other children why it was necessary for them to devote so much time to that one child, 59% did indeed consider that their other children felt left out. Surprisingly the neglect felt by the siblings did not seem to be a function of either the severity of the C.F. child's condition ($X^2 = 0.26$, d.f. = 2, $P \simeq 0.90$) nor of the total size of the family ($X^2 = 2.87$, d.f. = 5, $P \simeq 0.70$).

The question of whether and how this information and these feelings, would affect the behaviour of the well children

to their sick sibling, was rather a complex one. Certainly 72% of the mothers did suggest that cystic fibrosis was an intervening factor in the relationships between their children. In an effort to quantify the mode of this influence and the degree of its effect on the sibling relationships, mothers were asked about the behaviour of each of their healthy children, in turn, from eldest to youngest, towards the C.F. child. The scheme by which this information was collected is shown in question 277 of the mothers' first schedule and in Table III.F.(ii).

Initially all concessions made by the siblings were considered together, to provide evidence about the extent of the intrusion of cystic fibrosis into the children's relationships. However, the fact that the total scores obtained by each of the siblings ranged from 0 to 16 and showed a mean of 6.18 and a standard deviation of 4.98 reveals little until compared with control findings, although it is interesting to note that the children's sisters are rather more likely to modify their attitudes than are their brothers. (Mean score for girls : 6.60, for boys : 5.90). It is worthwhile to consider too, the separate aspects of the siblings' response. It was hypothesised that the reactions of older and younger siblings would be qualitatively different and the hypothesis was confirmed by the data presented in Table III.F.(iii).

Table III.F.(ii). To assess the Influence of Cystic Fibrosis on Sibling Relationships

	3	2	1	0
	very much	mod.	very little	not at all
(a) Does he give in more easily to N?	-	-	-	-
(b) Is he less aggressive than otherwise to N?	-	-	-	-
(c) Does he feel protective to N?	-	-	-	-
(d) Does he feel responsible for N's wellbeing?	-	-	-	-
(e) Does he feel jealous of attention given to N?	-	-	-	-
(f) Does he feel worried about N's illness?	-	-	-	-
(g) Does he ever talk to N about his illness?	-	-	-	-
	Total Score 00 - 21			

Table III.F.(iii) The Responses of Elder and Younger Siblings to the C.F. Child.

	Elder Siblings (N= 40)		Younger Siblings (N=32)	
	Mean Score	S.D.	Mean Score	S.D.
*(a)	2.6	3.5	0.3	0.7
(b)	2.6	3.6	0.2	0.7
(c)	3.4	3.2	0.6	1.0
(d)	2.5	2.4	0.4	0.8
(e)	1.4	1.6	0.7	1.3
(f)	2.5	2.7	0.4	1.0
(g)	0.6	0.8	0.2	0.6

* These letters refer to the items listed in Table III.F.(ii)

The results suggest that older brothers and sisters are more likely to respond, and to do so more strongly, in respect of the child's disease, than are younger siblings. Among the elder siblings, caring and tolerant behaviour responses predominate while among the younger children jealousy was most frequently and most strongly scored. It was very striking that few of the siblings of whatever age, ever talked to the child about his illness.

The question of whether or not mothers had explained the situation to the child's siblings was an important one. The associations between having had such an explanation from mother and giving in more easily to the C.F. child (X^2 = 14,2, d.f. = 7, $P < 0.05$).or feeling protective about him (X^2=14.9, d.f.=7, $P < 0.05$)

or feeling protective about him (X^2 = 14.9, d.f. = 7, $P < 0.05$) were significant at the 5% level, among older siblings. So, too were the reports of worry about the child's illness on the part of older siblings (X^2 = 14.3, d.f. = 7, $P < 0.05$) while for younger siblings, explanations seemed, if anything, to increase feelings of jealousy. This trend did not reach statistical significance (X^2 = 5.55, d.f. = 3, $P \simeq 0.2$) but in any case caution is required in interpreting findings concerning these younger siblings since several of them are still babies. The average age of the younger siblings in this group was 4:2 years.

In the final analysis mothers' responses proved utterly consistent with their original view and 28% of mothers described. all their other children as behaving quite normally to the C.F. child. However in 8 of these 11 families the siblings were all under the age of 5:0 years. Fathers' impressions of the responses of their healthy children to the C.F. child were marginally less favourable than mothers', with 25% of the 36 fathers recalling experiences of resentment of the C.F. child by his brothers or sisters.

The literature had suggested that these siblings would exhibit any of a variety of behaviour problems in order to detract attention from the C.F. child to themselves and so mothers and fathers were asked in a very general way about the kinds of problems which their other children, in their own right had presented. The parents' reports again failed to reveal behavioural pathology of the degree predicted by the literature. 12 of the 39 mothers did complain of temperamental difficulties in their other children, although only 5 of the 36 fathers were

aware of any such problems and the degree of agreement reached between parents was not high (X^2 = 2.37, d.f. = 2, P $\simeq$ 0.30). Nevertheless there were isolated examples of siblings feigning illness or otherwise seeking attention, in the manner described in the literature, and the relationship between the incidence of these problems and of the assessment of sibling resentment of the C.F. child was notably high (X^2 = 49.2, d.f. = 4, P < 0.001). It is only fair to add here that 14 of the 39 mothers and 11 of the 36 fathers in this group admitted that they got more pleasure from the C.F. child than from the other children in the family and the incidence of favouritism, expressed in this way, showed a relationship, approaching statistical significance, to the incidence of reported difficulties with siblings (X^2 = 9.31, d.f. = 4, P < 0.10).

If the children were disturbed in their relationships within the home, it was of interest to gather some information about their behaviour outside. This was particularly important in the view of the speculation in previous research reports, about delinquent tendencies among the siblings of C.F. children. For this reason Bristol Social Adjustment Guides for 'the Child in School' were given to the class teachers of all of the siblings in the group aged between 5:0 and 16:0 years. Guides were also completed for matched controls and the distribution of scores obtained, following Stott's Scheme as before, is given in Table III.F.(iv).

Table III.F.(iv) Distribution of Scores on Stott's Unract and Ovract Scales - For School Age Siblings of C.F. Children and their Controls

UNRACT	0	1	2	3	4	
Siblings	21	5	2	3	5	
Controls	29	2	0	2	3	0 : (near-) stability
						1 : mild reaction
OVRACT						2 : appreciable reaction
Siblings	19	4	2	5	6	3 : maladjusted reaction
Controls	29	2	1	4	0	4 : severely maladjusted

N (Siblings) = 36 : N (Controls) = 36

On the basis of the first analysis there was some evidence that the siblings of C.F. children were more likely to show disturbed behaviour patterns than were their controls. 42% of the siblings as compared with only 19% of the controls were described as showing some degree of under-reactive behaviour, while 42% of the siblings and 22% of their controls were said to exhibit over-reactive behaviour.

The mean scores obtained by the groups on these scales are shown, separated by sex, in Table III.F.(v). The mean scores also quoted by Stott (1971) are also given.

Table III.F.(v) Mean Scores attained by the Healthy Siblings of C.F. Children, and by their matched controls, on Stott's Scales of Unract and Ovract.

	Unract	Ovract
Brothers (N = 21)	M = 5.61; S.D. = 8.78	M = 15.0; S.D.=19.5
Controls (N = 21)	M = 4.76; S.D. =10.2	M = 2.71; S.D.= 5.37
Normal Boys	M = 3.05; S.D. = 3.66	M = 5.11; S.D. =6.60
Sisters (N = 15)	M = 3.46; S.D. = 5.79	M = 4.00; S.D.=9.80
Controls (N = 15)	M = 1.13; S.D. = 3.43	M = 1.93; S.D.=6.23
Normal Girls	M = 2.84; S.D. = 3.83	M = 2.71; S.D.=4.80

These scores suggest that over-reactive behaviour presents the more serious problem. It should be recalled that it is this mode of maladjustment which Stott (1971) views as the precursor of delinquent behaviour. Although in a sample of this size it is not possible to draw such dramatic conclusions, these findings are certainly sufficient to suggest a need for further research.

The whole question of further research which is now required in this problem, may be viewed in better perspective in the next Section where the results reported here, are interpreted in the light of the investigator's observations.

SECTION IV

INTERPRETATION OF THE FINDINGS REFERRING TO CYSTIC FIBROSIS CHILDREN AND THEIR FAMILIES

Here, we seek to qualify and to amplify, by means of verbal reports, those findings which were analysed numerically in the previous section. Those results will be discussed under sub-headings in this section, in the order in which they were first presented. However, the factual account of "The Family Setting", in Section III,A., requires little further comment, so its information will be included in the course of other areas of discussion, as appropriate, and this section will begin with a review of parents' experiences at the time of the first diagnosis of cystic fibrosis in the family.

We are also afforded the opportunity here, of comparing the findings of this study with those reported in the literature referring to children with visible handicaps. As far as these research reports allow, we shall endeavour to note similarities and differences in the findings and, thereby, to estimate the significance of the factor of visibility in the influence of the disease on the lives of the affected children and their families.

The Diagnosis of Cystic Fibrosis (Section III.B.1.)

The influence of cystic fibrosis upon the lives of affected families begins even before the disease is diagnosed. The characteristics of the undiagnosed fibrocystic infant are very distressing to parents, in particular, the common presenting

symptom of failure to thrive. Since the dominant function of the maternal role in the early months is to nurture, any failure in this respect may have a significant effect on the early mother-child relationship. Certainly, several of the mothers in this sample described feelings of self-doubt at this stage, reproaching themselves that their method of feeding their child was unsuccessful. It has been suggested that early difficulties may arouse feelings of hostility in the mother toward her child (McCollum and Gibson, 1970.) This was found to be rare in this study although in one instance, when the difficulties had been allowed to continue, the mother's despair did seem to be leading to such feelings.

The early symptoms of cystic fibrosis are not initially, readily distinguished from those of other more common infant ailments so it is perhaps not surprising that in the past, when cystic fibrosis was even less well known than it is today, long delays were reported in obtaining the correct diagnosis. Although, objectively, there is a trend to earlier diagnosis, as cystic fibrosis becomes better known, the degree of difficulty experienced by parents in trying to obtain the diagnosis does not seem to have shown a commensurate decrease, judging from parents' reports of more recent diagnoses. This may, of course, be a 'recency effect' that the more recent traumatic events are remembered more vividly than those of diagnoses made several years ago, and indeed this is a real hazard of using retrospective accounts as a source of information. Nevertheless, if we examine the reasons for the difficulties, it becomes evident that it is not only misdiagnoses which are troublesome.

There was a trend,for young mothers in particular, to be chastised by their family doctors for being too fastidious about

their babies' eating and bowel habits and in such cases, where the mother's inexperience of babies was blamed for her agitation, no action was taken. This source of difficulty was particularly traumatic for the mothers, and in the long-term, particularly damaging to the doctor-parent relationship. Although delays and misdiagnoses did, in any case, result in a mounting mistrust, of the medical profession among these parents, the blame that was attributed to the family doctors in these cases did not usually create a permanent loss of faith in doctors. These young mothers on the other hand, whose suspicions had been dismissed as unfounded, without any investigation, were more bitter. They tended not only to blame the doctor for his inattention but also to remove their family from his care to become patients of another doctor.

The difficulties did not always stop when the child was referred to hospital for an expert opinion. Mothers described difficulties here, in finding anyone who was willing to discuss what was happening to the child as diagnostic tests were made. Long delays or misdiagnoses at this stage could also be disturbing to the parents.

For almost a third of these mothers then, experiences at the hands of doctors during this prediagnostic period had a lasting effect, either to the good, or to the detriment of their subsequent attitudes to the profession. Although Kulczycki et al (1969) and others have suggested that the age of the child at the time of the diagnosis is critical to the family's later attitudes and Debuskey (1970) has asserted that "under all circumstances the first task is to establish a medical diagnosis as carefully and swiftly as possible", the views of the parents included in this study would

suggest that it is not only the time factor, of delay in obtaining the definitive diagnosis, which is so traumatic for them, but also that the course of events during that prediagnostic period, is significant. Good management of the C.F. child and his family should, then, like the influence of the disease itself, begin even before the diagnosis is made.

Once the differential diagnoses have been excluded the physician has an obligation to inform and advise the family of the nature of the disease and its likely course and outcome. Clearly the communication of this diagnosis cannot be stereotyped and must be modified by the uniqueness of the personalities involved. Nevertheless some common factors have emerged from the study which may act as a framework wherein this highly personal communication can most effectively be given.

All too often, in the cases in this study, the diagnosis was communicated to mothers alone, and in only one third of the cases had the diagnosis been given to both parents together. Although a few parents did describe feelings of momentary relief that their child did, at least, have a disease that the doctors knew how to treat, the predominant response to this communication was, understandably, one of shock. In spite of apparent intellectual acceptance at the time, most of the parents, particularly mothers, reported having retained little of what they had been told on this occasion.

It was this shock reaction and the selective forgetting or remembering of aspects of the diagnosis communication which made it so distressing for mothers to be told this news while they were alone. Several mothers described their nightmarish journey home afterwards and their subsequent inability to explain

the diagnosis to their husbands. Although it may not always be possible, or easy, to arrange for both parents to be present, this is clearly the optimal situation for the communication of this diagnosis.

Clearly, there is a fine balance to be struck in the communication of a diagnosis of cystic fibrosis, between an overemphasis on the prognosis, in a way which leaves little room for hope, and, an excessive delicacy which fails to convey the gravity of the situation. The problem is to find the approach by which this balance may best be achieved. On the whole, parents, particularly fathers, appreciated a direct approach which prepared them for 'the worst' but at the same time pointed out the means by which modern medicine could hope to control the situation. They were, nevertheless, very sensitive to approaches which they described as 'blunt' or 'tactless'.

Since shock precluded a proper appreciation of the diagnosis at the first telling, it was clearly important that there should be repeated opportunities for parents to acquire further information on later occasions, yet only half of the parents in this sample had had subsequent discussions with the diagnosing physician. The responsibility for this lack of discussion lay fairly equally with doctors and parents. Usually the diagnosis was first communicated to the parents by the consultant paediatrician in the hospital to which the child had been admitted. In a number of cases in the past, this doctor did not seem to have been readily accessible to parents after this time. In an equal number of instances, however, parents admitted that the doctor had said he would be available if they

wanted to see him but that they had felt afraid to disturb him. In view of the parents' emotional difficulties at this time it would seem that the onus for arranging some form of follow-up discussion should lie with the physician, for there can be little doubt of the need for such opportunities for repeated explanation of the diagnosis.

As for the first communication of the diagnosis, so for the subsequent encounters, there can be no hard-and-fast rules of procedure. Parents described a gamut of emotional reactions to the diagnosis and it seemed that it was helpful if they could discuss with the doctor not only the objective facts of cystic fibrosis but also their subjective feelings about these facts. A number of parents reprted that their distress had been somewhat alleviated by discussions of this kind and there was a suggestion of a halo-effect extending from the tone of experiences of this time to the tone of their later attitude to that doctor as a person, which could aid or hinder their subsequent relationship with him.

As the initial shock wore off, parents described feelings of resentment, "why my child?" or of disbelief, "it can't be true, they must have made a mistake," in response to the diagnosis and, in some cases, of blame or guilt, in reaction to the genetic basis of the disease. Some mothers also remembered feelings of anxiety about minor indistretions committed during pregnancy, which they feared might have caused the child's condition. Skilful management of the diagnosis should be able to alleviate these cources of distress but it is doubtful if it could, or even, if it should, interfere with the predominant 'anticipatory mourning'

reaction which sets in as the implications of the diagnosis become more realistically appreciated.

In this period of anticipatory mourning parents describe distress, grief, depression and disturbances of appetite and sleep in response to the threatened loss of their child. However, it seems that it is the passing of this aspect of parents' response which acts as the impetus for information-seeking behaviour and for active involvement in the child's treatment, both of which serve to dissipate parental anxiety and to restore the parents' confidence in their ability to care for their child. The parents of these children needed a great deal of support and understanding to help them through this most distressing period.

Although parents were asked what information they had, in fact, been given it was very difficult to ascertain what they had been told at the time of the diagnosis, for a number of reasons. The retrospective nature of these accounts allows both proactive and retroactive interference to operate so that what has subsequently been learned, since the diagnosis, may now be confused with what was originally made known to them then. At the same time, the defence mechanisms acting at that time, and subsequently, may have left parents with a very biased impression of what the doctor really said. Nevertheless these questions allowed an assessment of the information which parents had about cystic fibrosis at the time of the enquiry.

Parents were quick to point out at this juncture that the information had taken a long time 'to sink in' but, by the time of the enquiry, most of them knew that their child's condition was inherited and that it was unlikely ever to be cured.

Although only about half of the parents claimed to have been told about the likely outcome at the time of the diagnosis, it seems more likely that these responses reflect the operation of their defences. The need for doctors to inform parents of the outcome may be appreciated in the reactions of the few parents who had clearly not been told of the uncertainty of the prognosis at the time of the diagnosis. Their indignation and distress on being exposed to this information at a later date may not always be very rational, but it can be damaging to the relationship with the doctor thereafter, as the mother wonders what other information has been kept from her.

Understandably, since they had not always been included in the communication of the diagnosis, fathers' information was not always the same as their wives. Although the mothers did try to explain the disease to their husbands, the effect was not as satisfactory as when the fathers had themselves had had an opportunity to discuss the matter with a doctor. When the situation was unclear, new information from outside sources e.g. the media, could be interpreted differently by each of the parents in a way which, it was found, could lead to family tensions. Similarly, both parents needed to be armed against the disbelief of their relatives and friends whose palliative words could undermine the faith of the doubting parent in the diagnosis given by the doctor.

Although more than half of all the parents interviewed did feel that someone had taken trouble over explaining the diagnosis to them, less than half of the parents expressed complete satisfaction with the way in which this had been done. A number of points from these parents' interviews may be helpful in summarising

their views on how the diagnosis of cystic fibrosis might best be managed:

1. The diagnosis should be given to both parents together, by the consultant physician if the child is in hospital (as is most likely) and by the family doctor if the child is at home (rare, nowadays.)
2. The diagnosis should be discussed in a place where privacy is assured and at a time when freedom from interruption can be guaranteed. Clearly too this time should be when the diagnosis has been confirmed.
3. The parents appreciated a simple and honest explanation which did not confuse them with medical terminology or distress them by its bluntness. They welcomed information about the positive aspects of therapy which left them room for hope but accepted the need 'to know the worst.'
4. There was a need for repeated opportunity for questions to be asked and for feelings that hinder the acceptance of the diagnosis to be exposed. The physician is in a better position than the parents are, to initiate such opportunities for further discussion.
5. In the ideal situation, the parents are allowed some time to come to terms with this diagnosis before their child is discharged from hospital into their care.

Since information seeking behaviour was found to be a common development in parents' reactions to the diagnosis it was agreed by most parents that it would have been useful for them to

have had some written information to take home with them after the verbal communication of the diagnosis. This would have enabled them to check the accuracy of their recall of the information given and might have given them more confidence to ask questions. Parents clearly do experience difficulty in learning about cystic fibrosis from other sources at this time (Table III B (vii)) so it is important that the diagnosing physician provides as much information as is required, as unambiguously as possible.

Parental Comprehension and the Genetic Implications of Cystic Fibrosis (Section III B.2)

In spite of all that had occurred since the first communication of the diagnosis to them and in spite of their key role in the treatment of their fibrocystic children, parents' understanding of cystic fibrosis, as tapped in this study, was not particularly good.

Certainly parents who showed a higher level of intelligence and those who had had more years of formal education were more likely to have grasped the main facts about the disease but it was significant that fathers who were not included in the communication of the diagnosis at the outset were unlikely ever to achieve a good level of understanding of cystic fibrosis thereafter.

It was a particular concern of the study to learn of parents' response to the genetic basis of cystic fibrosis for this response has important practical repercussions in the family decisions which are then made.

In the first instance, the study revealed a significant lack of knowledge of the genetics of cystic fibrosis among these parents and a number of causative factors were identified.

It must be assumed that some account of the genetics of the disease was given to the parents at the time of the original diagnosis, although none of these parents had been referred for genetic counselling by a specialist. Clearly, in several cases, the explanation was not effective. Emery (1973) has already pointed out that a major obstacle in genetic counselling is public lack of knowledge of basis biology and genetics. The ordinary terms of the genetic vocabulary are often completely unfamiliar to parents. Semantic confusions were evident too in the interpretations placed even on more familiar words. For example, the adjective 'inherited' was often thought to apply to diseases passed from parents to children but contained within those two generations whereas the term 'hereditary', did convey that which was passed on through successive generations of the family. Language problems then clearly do contribute to the parents' poor understanding of the genetics of this disease.

As for the understanding of the nature of the disease, so again, it was found that parents of higher intelligence and those who had had more years of formal education were significantly more likely to understand the genetic risks. It is interesting in this context to note that the explanation given to parents of genetic risk factors tends to depend heavily on the parents having some concept of 'probability'. It has already been observed that probability is a developmental trait and that individuals differ in their ability to achieve this competence. (Leonard et al, 1972; Davies, 1965.)

Psychological defence mechanisms e.g. denial, also acted as barriers to comprehension. A few mothers (4%) completely refused to accept cystic fibrosis as an inherited disease in spite of all evidence and argument. A less extreme form of defence, avoidance, was demonstrated by the fact that although a number of parents felt that they did not fully understand the inheritance of the disease, not all of them felt motivated to seek out further information nor indeed would they all welcome such information being made available to them.

Thus lack of education, low intellectual ability and the psychological defence mechanisms are seen as the major barriers to parents' understanding of the inheritance of cystic fibrosis. Nevertheless, some responsibility must lie with the diagnosing physician for including his explanation of the genetic basis of cystic fibrosis, in with a welter of other information about the name of the disease and its likely course and outcome, and all at a time, as we have said, when too often only the child's mother was available to hear. The parents' lack of understanding of the genetics of this disease then becomes understandable when it is recognised that the only form of explanation offered was, so often, given at the time when they were suffering from the shock and distress of learning of their child's poor ultimate prognosis.

Since we were concerned with the stresses imposed on parents by the disease it is important to give thought here to parents' emotional reactions to the information that cystic fibrosis is an inherited disease. Lack of full understanding of the inheritance of the disease can undoubtedly contribute to the parents' unhappiness. (Section III.D.2) Where, for example, it was

believed that the affected child's condition originated solely in the spouse's pedigree, then feelings of anger and resentment were more likely to occur. A few parents, who knew that no such disease had ever occurred in their own families, assumed that it had been inherited from their spouses, but, happily this misunderstanding was unusual. Much more common as a cause of unhappiness among parents was the feeling of guilt or responsibility for the transmission of the disease to the child. This feeling was particularly troublesome to those fathers whose wives had had normal children by a previous marriage. Parents who had, themselves, a medical history of chest complaints, also seemed more likely to express anxiety on this point while parents who had a sound understanding of the genetics of cystic fibrosis were less likely to report feelings of guilt in this connection.

It was significant that the parents disclaiming feelings of guilt often attributed their outlook to physicians who, in their case, had explicitly explained that it was inappropriate for them to feel guilty. There seemed then, to be further grounds for suggesting that the parents' burden of distress could indeed be lessened if greater care were taken to explain to them the mechanism of inheritance of cystic fibrosis, and to discuss with them the irrationality of guilt feelings.

It is of interest now to consider parents' behavioural response to this aspect of the diagnosis, in terms of their family planning decisions. (Section III.B.2)

Only 36% of the parents in this study felt obliged to have fewer children than they had originally intended, because of the risk of conceiving a further child affected by cystic fibrosis.

In an equal number of families a decision to limit the family size was taken simply because the desired number of children had already been born. This finding showed an interesting contrast to Burton's report (1972) of the situation in Northern Ireland where 70% of the parents gave cystic fibrosis as the reason for the limitation of their family size. In this context, Leonard et al's point (1972) that "the strong relation between parents' conception of the burden of the disease and their reproductive attitudes suggests that burden, as distinct from risk, has a large role in decisions about future childbearing," is highly relevant. The Irish families in Dr. Burton's sample had had a quite different experience of child mortality and morbidity from cystic fibrosis than had the Scottish families. More Irish children had died, and, unlike the Scottish deaths, many of these children had survived for several years. Thus, these findings would seem to endorse the views of Leonard et al. by suggesting that the experience of morbidity associated with the disease has more influence on parents' decisions than the knowledge of the likelihood of early death.

In a sense the validity of this concept of 'burden' was further strengthened by the observation that when the original decision to limit the family size, because of cystic fibrosis, was made easily and without conflict, the parents maintained this position. Where, however, the parents had originally accepted advice or had been influenced in reaching their decision, they were more likely, later to review the situation in terms of their own subsequent experience of the disease and to base further decisions on the observation of the burden which would be imposed upon them by the birth of another fibrocystic child.

These findings may be useful in the approach taken to the subsequent genetic counselling given to these parents.

It was disappointing to find that the outcome of family planning did not always match the parents original intention. The incidence of accidental conception was rather high, indicating a need for more and better contraceptive advice. As a result of these findings, parents attending the Cystic Fibrosis Clinic in Edinburgh are now offered Family Planning advice and a suitable appointment is arranged for them at a Family Planning Clinic.

Parents' Attitudes to General Practitioners (Section III.B.3)

In a number of cases in this study, parents' confidence in their family doctor was shaken by his failure to diagnose the child's disease himself or to recognise, sufficiently promptly, that the child required specialist attention. As we have already indicated, in a few cases, this was generalised into a total lack of confidence in his ability, and the family changed its doctor. The majority, however, maintained contact with their family doctor and 60% of the families in the study still consulted him about their cystic fibrosis child, in spite of their regular visits to the out-patient clinic.

It was interesting that those who were frequently brought into contact with their family doctor were not only the young children, who do tend, even without cystic fibrosis, to require frequent attention for minor ailments, but also the children of manual workers. There was a tendency for the mothers of this latter group to experience social class barriers to their communication with hospital doctors and several of them described

their general practitioner as being more approachable and as having a better understanding of their problems than the clinicians.

Although as many as three out of every four mothers reported that their G.P. took an active interest in their child's case, it was clear that the contribution that he could make to the child's care was often hampered by his lack of specialist knowledge. The mothers knew this too, and although many of them welcomed the doctor's interest, and some relied on him for emotional support, most mothers expressed some reservations about his competence to advise about the child's cystic fibrosis.

Mothers' relationship with their family doctors was further complicated by a rather strange feature of the treatment system. Although the children's cystic fibrosis was reviewed at the hospital out-patient clinic, the clinic could only advise on home treatment, prescriptions were not given out there. Thus, a letter had to be sent from the clinician, who had seen the child, to the family doctor, commenting on his findings and recommending a course of treatment, for which the family doctor would then be expected to write a prescription. This practice is general to the operation of most out-patient clinics, and not confined to those dealing with cases of cystic fibrosis. Nevertheless in these cases, treatment which is urgently required may be held up, and delays and confusions were not unknown, diminishing mothers' confidence still further. The arrangements made for mothers to collect these prescriptions from the G.P.'s were, sometimes, a further source of difficulty in the doctor-

parent relationship, since the interests of the doctors'and the mothers' convenience were not always compatible.

Certainly, the family doctors had a very useful part to play, particularly in the early years while mothers were coming to terms with the diagnosis and learning competence in the care of their C.F. children. However the existing workload of many family doctors, their lack of specialist knowledge about the disease and the sometimes impoverished communications between the doctor in the community and the doctor in the hospital, all mitigate against these doctors in their attempts to fill that role satisfactorily.

Clinics and Clinic Doctors. (Section III.B.3.)

Most mothers bring their children from a distance of fifteen miles or more to attend the C.F Clinic. More than a third of all the families who come have to rely on public transport and, for some of them, this creates financial difficulties. More than one fifth of them travel by ambulance for which they do not have to pay, but which may involve them in a much longer time commitment, than public transport would do, since a door-to-door service is provided for as many people as possible during the journey.

The time which mothers had to spend, away from home, varied from more than an hour to more than six hours. For mothers of young children this could be an arduous journey, while for older children, it required an afternoon off school, in many cases. Mothers who had other children had also to make arrangements for them to be looked after, during their absence, or else they had to cope with bringing several children to the clinic.

If mothers are to be asked, to tolerate such a commitment in time, money and effort, and to attend the clinic on a regular monthly basis, then these requests must be seen by them to be justified.

Mothers did not complain about the time they spent waiting to see doctors at the clinic, partly because the delay was generally agreed not to be excessive, but also because the mothers were happy for their C.F. children to have time to play with toys, so that the clinic did not seem too strange or frightening to them. The mothers did complain that the time they spent in the consulting room was not long enough. The large majority of the mothers estimated that this time was 15 minutes or less, and they pointed out that since it was unusual for them to see the same doctor on consecutive visits, not all of this time was of benefit either to the children or to them. Several mothers, admittedly of generally fitter children, said that they attended the clinic primarily for the benefit of the doctors, to help to add to their knowledge of cystic fibrosis. Several others, usually those who had a long, inconvenient or expensive journey to make, expressed the view that the family doctor could just as well supervise the child's condition at much less trouble to the family. The representatives of these two points of view, and especially of the latter viewpoint, tended to be irregular clinic attenders, and it was these attitudes which seemed to underlie the high degree of association reported between the family's distance from the clinic or their financial situation and the regularity of their clinic attendance. (Table III.B.(xxii)) It was interesting to observe that there were definite personality factors involved in clinic attendance. This suggested that, whatever the circumstances,

the mother who was "anxious to do the right thing, attentive to practical matters and subject to the dictation of what is obviously possible" was by her very nature more likely to bring her child regularly to the outpatient clinic. Conversely, the less conventional mother who was unconcerned over everyday matters was in any event less likely to be a regular attender.

Clearly, then, the course of events during the time that the mother spends in the consulting room is critical to her evaluation of the worth of regular clinic attendance to her and to her C.F. child.

Mothers held two main expectations of medical supervision of their child's disease, in addition to the provision of medical treatment. They looked to the clinic and clinic doctors for information and for a degree of emotional support. It was evident from their opinions that the existing situation was not entirely satisfactory in meeting either of these needs.

A major issue was that of communication. More than half of the mothers in the sample were dissatisfied with this aspect of their relationship with clinic doctors and, where this was the case, their attitudes to more general aspects of the clinic tended also to be less favourable. Mothers complained of inadequate information about their child's progress, about the treatment prescribed for him and about the clinical tests which were, from time to time, conducted on him. For their own part, some mothers reported feeling inhibited in asking questions by the fear of sounding foolish or by social class barriers, while others were inhibited by the presence of others in the consulting room. Where questions were not asked, naturally none were answered, and the communications between some of these mothers and their doctors were certainly impoverished.

It is only fair to add, however, that for the remainder of the mothers, almost half, no obstacle had been allowed to stand in the way of their understanding of the child's condition: "I just used to keep a list of all the things that puzzled me between clinic visits. I'd write them all down and take them along so I wouldn't forget. I don't think they (the doctors) really liked me sitting there asking so many questions at first, but now it's all right. I'm not afraid to ask anything I don't know about and they are very good about explaining it to me." It is significant, however, that positive attitudes tended to be expressed by mothers who acknowledged, but had overcome, the inhibitions against asking questions of the clinic doctors.

The second main point in mothers' expectations of their clinic attendance was that it would prove a source of emotional support to them. This desire was reflected in the large proportion, (about 70%), of mothers, who said they would prefer to see the same doctor each time they attended the clinic. Although there were practical justifications for this request e.g. the mother would not then have to reiterate the same information each month to a different doctor; there would be a better assurance of continuity of medical care for the child, the reasons given by the majority of these mothers were more emotive, e.g. the need they felt to betreated as people, rather than as cases of cystic fibrosis. By the same token, two-thirds of this group, almost half of the total sample, said that they preferred their child to see the consultant. Rationally, they acknowledged that other doctors had to learn about cystic fibrosis and that the general standard of care was carefully supervised and uniformly high irrespective of which doctor they saw, but the psychological

significance of the fact that the consultant had examined the child was tremendous, in its power to reassure mothers.

In fairness to the system of clinic administration, we must point out that, over the eighteen months that this part of the study was in progress, there were only three doctors who regularly saw these children, although one was replaced by a fourth early in the study, and that they did make every effort to get to know the families with whome they came into contact. Continuity of care was sought, by these doctors coming together at the end of the clinic to describe the cases they had seen and the treatment they had recommended. It was, therefore, difficult to see how the system could continue to work so efficiently and yet provide any further support for these families within the existing framework.

The points made by mothers which could well be noted within the eixsting system, may be summarised as follows:

1. There is a need for the doctors at the clinic to provide mothers with more information and explanation.
2. There is a need to create an atmosphere and time in which mothers feel they can ask questions.
3. Although it is not routinely possible, there should be more ready provision made for parents to see the doctor of their choice, in cases of necessity, and for them to be able to see him without having either the C.F. child or any others in the consulting room, if they so desire.

Thus, although the mothers show by their behaviour that they are satisfied with the system of medical supervision offered

by the clinic, 80% of them attend on at least a fairly regular basis, their verbal reports suggest that there is still room for some improvement.

Treatment (Section III.B.4.)

The responsibility which these mothers have to take for the treatment of their C.F. children is indeed a heavy one, although in many cases it is less now than formerly. There is a number of medicines to administer, usually several times a day. The mother is required to keep supplies of these in stock at home and to ensure that correct dosages are given at correct intervals of time although in some cases the exact details may be left to the mother's discretion. Formerly the mother was required to see that the child spent some time in a mist tent, once or twice during the day and during the night, in preparation for his physiotherapy although nowadays most of the children have this postural drainage without the preceding mist therapy. These responsibilities placed on the mothers were in addition to the normal ones, of providing an adequate standard of nutrition and hygiene, two tasks which may be made more difficult in the case of C.F. children.

Undoubtedly mothers find physiotherapy, the most time-consuming, and to them, the least natural of these tasks, the most troublesome.

While their C.F. children were infants, mothers reported little difficulty in the administration of treatment, although at the age of tantrums, around 4:0 years, a few problems did arise. The difficulties, at this age, were usually those of the mothers. Mothers naturally, expressed a lack of confidence in the handling

of their infants in the early years after the diagnosis. They were uncertain about which manifestations represented problems requiring immediate medical attention and which were transient or normal; where did the influence of cystic fibrosis end and the normal infant begin? Mothers' unfamiliarity with the treatment they had been asked to administer created additional difficulties. They described their valiant efforts to make the treatment palatable to their infants, their consternation when, inevitably they made mistakes, and a host of other minor problems of the domestic aspects of the management of the treatment of the C.F. child.

Although the mothers' difficulties decreased, as they became familiar with the treatments and as they became more confident in the handling of their children, other problems then began to emerge. As the children grew older, particularly as they went to school, and as they approached adolescence, they tended to resist treatment, complaining both of its time-consuming intrusion into their lives or of the embarrassment it caused them. At the younger stage, mothers could enlist the help of school authorities in ensuring that their 5-8 year olds did, in fact have the medicines prescribed although even at that age much depended on their powers of explanation, to the childrens satisfaction, that the treatment was all necessary. For the adolescents, good communication with a mentor, whether parent or other seemed to be the only way by which a positive attitude to therapy could be restored.

Since rigorous adherence to the treatment regime prescribed, is the only means that modern medicine can offer to cystic fibrosis

children, by which their condition can be controlled, it is vital that a positive attitude to therapy is maintained throughout among parents and children alike. Mothers' views expressed in the course of this study, suggested that some assistance was required in the domestic matters of administering this treatment, particularly in the early years. It also became clear that the mothers needed to know more about the medicines they were giving the child, and why, both for their own satisfaction and in order that they could reasonably enlist the cooperation of their children.

Agents of Help (Section III.B.5.)

A fairly clear picture had already emerged of the kinds of help that these mothers seemed to require, but, before any recommendations could be made, it was also necessary to review the explicit needs which they themselves described, and the ability of the existing facilities to meet these needs.

It was clear that some of the mothers, particularly those with very young children or with large families, had more responsibilities to meet than they could reasonably cope with in an average working day. To help them in their tasks some mothers expressed a need for domestic help to free them to spend more time in the care of their C.F. child, others expressed a need for help with the time-consuming physiotherapy, in order to free them to spend some time on their homes or other family members. The needs of these mothers were fairly long-term, measurable in months, or even years, of help required. Another section of mothers explained that there were periods of shorter duration e.g. when the mother was ill or pregnant, when they were unable to meet their

responsibilities in their usual fashion. If the C.F. child's health was not to suffer during these times some help was required, particularly, again, in the administration of physiotherapy.

The existing social services were not geared to meet these needs and a more specialised investigation would, no doubt, be necessary before any attempt could be made to make any such provision of domestic help or of help in physiotherapy. As an expedient, it might be suggested to families expressing such needs, that the physiotherapist at the hospital would be willing to arrange to teach other family members to give this therapy correctly, in order to ease the burden on the child's mother. Fathers and other family members in several families had already learned how to administer this treatment and were able to be of some help to mothers in this respect, at least in emergencies, if not in a routine way.

The needs for information and for emotional support and reassurance had already been observed in relation to mothers' expectations of the medical supervision provided. Their need for this assistance to be made available on a domestic level, was apparent from the mothers' experiences of programmes of home treatment for the children. It was clear that the existing social service agencies, e.g. Health Visitors, completely failed to meet the rather specialised needs of these mothers. Although domestic crises arose in most of these families from time to time, when the mothers longed for someone to turn to, the need for a supporting service in the homes was seen to be most acute in the early years following the child's diagnosis.

As a result of these findings a new dimension was added to the system of care extended to these families. Begun initially on an experimental basis, a trained nurse was made available to meet the needs of these families, who children were patients of the Royal Hospital for Sick Children. This nurse was familiar with the pattern of in-patient and out-patient care in the hospital and she participated in ward rounds and clinics concerning the C.F. children, thereby becoming familiar with the mothers and their children. She was also aware of the patterns of care in the community and could provide a liaison with family doctors and Health Visitors. However the greatest value of her function was that she was free to visit families in their homes, at their request, to provide for those needs that had been unfulfilled by the previously existing system.

This nurse then, had competence to advise on the domestic matters of treatment which worried these mothers and yet which they felt to be too trivial to ask the doctors about. She could provide information and reassurance in an unequalled way, by being less awe-inspiring than the hospital doctors, more authoritative than the family doctor and, in some cases, simply, by being a married woman. For queries which required only a brief response, parents were then able to telephone this nurse in the hospital to obtain advise, where previously they might have delayed, fearing to trouble the busy doctors. Since the nurse was based in the hospital and able to maintain a close working relationship with the consultant paediatrician, the policy regarding the care of any individual child maintained its consistency. The same nurse was then available to provide a welcome personal touch to the child's subsequent visits to the clinic and to his hospital admissions.

In the early stages of this home visiting service it was necessary for the nurse to call on mothers to discuss her proposed role and to introduce them to its possibilities. Since the needs were, as anticipated, greatest among the mothers of children who had been recently diagnosed, the nurse was involved in providing an extensive supporting service in these homes. By observation, her visits were very successful in meeting the needs of these mothers, but, since the pattern was still emerging, there was no justification for including these mothers in this study. It would be interesting, as a validation of the usefulness of this extension of the system, to compare the views of these mothers, at some time in the future, with those of the mothers in this study, who had to cope with their cystic fibrosis children without such assistance.

Hospitalisation (Section III.B.6.)

Mothers' attitudes to their children's hospital experiences were much harder to elicit and, on the whole, much less illuminating, than the attitudes which they expressed about other aspects of the children's medical supervision. The standard of hospital care provided for these children is certainly high, but an additional reason for mothers' tacit acceptance of hospital procedures may simply be that they perceive the child's urgent needs for treatment beyond that which they themselves can administer and are relieved and grateful that the hospital services can take over the responsibility.

Although there were some minor criticisms about domestic arrangements within the hospital and, again, some observations

about the difficulty which some mothers had experienced in obtaining information about their child's progress or about the procedures to which he was subjected, on the whole, the views expressed were positive in their tone.

Since the study had set out principally to describe the behaviour of the children in their natural environment it is ill-qualified to make more than a few very general observations here. A further obstacle to generalisation, on the basis of the findings of this study, was the very widely varying hospital experience of the children in the sample. It might be expected that the older children would have had more hospital admissions than younger children, but this ignores the all-important factor of the severity of the child's condition. Whatever their age, children whose health was graded C on Dr. McCrae's scale were more likely to have had considerable in-patient experience than were their contemporaries whose condition was graded A or B.

In the case of young C.F. children (under the age of five years), mothers described very characteristic behaviours indicative of the children's distress at being left by their mothers. Mothers, quite reasonably, assessed these children to be rather too young to benefit from any verbal explanations and they described their efforts to minimise the children's distress in terms of their staying with the child as much as possible. Although there is no accommodation for mothers in the Royal Hospital for Sick Children, there are no visiting hours and it was possible for mothers to spend as much time with their hospitalised infants as their other family commitments would allow.

Nevertheless, these young children invariably protested vigorously at being hospitalised, and were highly likely to show

behaviour disturbances on their return home. None of the children in the sample had been hospitalised for long periods at this age so that the deprivational effects of the impoverished experiences of hospitalised infants were not a problem here. Although these young children attended the out-patient clinic and had the opportunity to become familiar with the staff, they seemed to present a similar degree of protest to each hospitalisation at this stage.

It was more easy to prepare the older children for hospital admission and several of the mothers did like to do so. Several mothers said their children had come to recognise in themselves when hospitalisation was necessary. The older children were less likely to exhibit protests in the manner of the infants, and indeed some of them who had been in hospital several times seemed to accept hospital admission with stoical resignation. Nevertheless, there was a strong element of anxiety attached to the children's stays in hospital and mothers often described the children as being relieved to be home. Mothers of the oldest children in the sample, who had been in hospital several times, had rather a different view. They felt that their children attended closely to all that happened or was said, in the ward and that the information which they gathered in this way, or the interpretation which they gave to these events could have a disturbing and depressing effect on their morale.

This introduced a rather sensitive problem in the care of these young people. The organisation of the out-patient clinic was such that C.F. families who did not already know each other were unlikely to become acquainted. Within the confines of the

hospital ward the situation was rather different. Mothers with time on their hands were more likely to talk to other mothers visiting in the ward at the same time. In this way, C.F. mothers could meet and, if the opportunity arose through subsequent clinic and ward encounters, both mothers and children could become acquainted. The likelihood of this event having occurred was clearly greater in the cases of older children, in poorer physical condition who had had frequent hospital stays. These relationships then tended to be maintained, and although they could clearly, have a positive effect, in helping the mothers and the children to feel that they were not alone with their problems, they could also have a very deleterious effect, if the identification went too far, for instance, when one of these children encountered severe difficulties or died.

Considerable sensitivity had been shown in the handling of young people with cystic fibrosis, who had presented emotional difficulties, for these reasons, in the hospital ward. Experience to date suggests no easy answer, but it is clear that this aspect of the care of cystic fibrosis children is one which merits some further attention.

Financial Pressures of Cystic Fibrosis (Section III.C.1.)

For the National Health Service patients who constituted this sample, cystic fibrosis did not create a serious drain on their financial resources. That is to say, the families, as whole units, were not deprived of essentials of living, e.g. food, clothing or shelter, in order to sustain the C.F. children among them. Nevertheless the economic issues of having a cystic fibrosis child could not be overlooked, for it was clear that financial pressures

could only exacerbate an already stressful situation for parents, and that mothers who had continually to worry about the family budget were less able to cope with the care of their C.F. children.

Clinic and hospital visits could be costly but there is provision within the social services, for mothers in need to recoup these losses. However, more than a third of the mothers in the sample described additional expenditures caused by the C.F. child. In the case of food and clothing it was clear that the family budget was being stretched to provide a better standard of living for the C.F. child than the family could otherwise sustain, at the mothers' own instigation.

The effect that the C.F. child had in causing the reorganisation of the priorities of the family budget seemed most evident when the child's health was poor, or only moderately good and when the family's income fell in the middle range. Families who were, at least, 'comfortably off', reported no financial difficulties and poor families were satisfied if they could feed the family, pay the bills and keep out of debt, but families in the middle group strived for luxuries, e.g. cars or telephones which they saw would be of benefit to the C.F. child. When families reorganised their budget in this way, for the good of the C.F. child, whether in food, clothing or extras, then other things had to be sacrificed, and it seemed to be most usually in these cases that C.F. was described as placing a strain on the family budget.

Where mothers felt unable to work because of the needs of the C.F. child, or where fathers had lost work and, hence, pay over the child, financial stresses were again more likely to be reported.

Several mothers mentioned the earlier source of economic

drain they had encountered in mist therapy. Although the mist tents had rarely caused structural damage in the homes of this sample, they had taken a heavy toll in beds and bedding, and in some cases in interior decoration, and mothers were relieved when this mode of therapy was discontinued.

Pausing for a moment on the topic of housing, it was recorded that the cystic fibrosis children were not described by their mothers as requiring anything special in the way of housing accommodation, so that none of these families had had to modify their homes. The children did have the needs of any child, in that their homes should be structurally sound, that they should have adequate facilities for the families' needs and, preferably, that they should have a safe play area or garden for the children. On this basis it was understandable that the families were not routinely given any special concessions in Local Authority Housing Departments. Nevertheless, these C.F. children are particularly likely to suffer if the home is damp, overcrowded or lacking in play facilities. Their health, may, at the other end of the housing scale, make central heating unsuitable in some cases, so that some concession in the Housing Departments' usual procedure for the allocation of houses may need to be sought. In the limited experience of families described in this sample, Local Authorities seem to have been willing to help in this matter.

Social Isolation of C.F. Families (Section III.C.2.)

In common with the findings of Turk (1964) this study did offer evidence that cystic fibrosis families were likely to give up having family holidays. However, further discussion revealed that only one third of the families, who had not had a family

holiday since the C.F. child was diagnosed, attributed this lack specifically to cystic fibrosis. Families with very young children, with limited budgets or with strained marital relationships between parents were more likely to give these as reasons for lack of holidays.

The socially isolating effect of cystic fibrosis on family activities, where it was described, seemed to have evolved for two main reasons. Firstly, even before the children were diagnosed, mothers reported a feeling of shame over their children's sickly appearance and in particular, an acute embarrassment about the strong smell which emanated from the children's nappies. This embarrassment could extend into the post-diagnostic period, for as long as toilet problems persisted. Since some of the children were themselves distressed by this aspect of their condition and unwilling to use toilets in homes other than their own, respect for their feelings could, in this way, affect the family's activities for a number of years.

The second factor in the social isolation of these families was often the mothers' anxiety. Mothers who were afraid of leaving their C.F. children in the care of others or who were anxious about taking the child and his treatments away from home, were clearly imposing a degree of isolation upon themselves, especially if this anxiety extended to a restriction on incoming visitors, lest they be harbouring germs which could endanger the C.F. child's health.

Social isolation in Turk's terms (1964) was not very common nor where it did exist, was it severe, among the families in this sample, but it seemed a great pity if families who were so deserving of holidays or outings, could not have them. The

C.F. Trust helps in this problem by providing large caravans, on good, permanent sites, with all amenities, in a few places in the country, for holidays for these families. However, the ideal holiday for these mothers would clearly be one where the responsibility of the child's treatment could be taken from her shoulders for a time.

Family Relationships (Section III.C.3.)

The main difficulty, in conducting any study which purports to assess the influence of, for example, cystic fibrosis on family functioning, is one of knowing where the base line is. How did the family operate before, or, how would it operate if the child did not have cystic fibrosis? Some consideration must be given to these imponderables if we are to hope to describe, fairly, the impact of the disease on the family.

With this difficulty in mind, the decision was made that the investigator should avoid making value judgments and that it should be left to the families themselves to convey just how intrusive a presence they found cystic fibrosis to be. It is on this basis that the influence of the disease on family relationships, within the families in this sample, to describe.

The influence of cystic fibrosis in the child's relationships with others was examined first. Naturally, relatives and friends are concerned about the child's condition, but it is the way in which they show this concern, particularly when the child or his well siblings are present, which can be disturbing. Happily more than half of the mothers had had no difficulties in this respect or had found it easy to explain to relatives and friends that they preferred not to make an issue of the child's condition. In the

remainder of cases the effect was usually evident in pampering. Not surprisingly when the C.F. child was too frequently the recipient of treats, or the centre of attention, sibling rivalry would be aroused. Occasionally the concern of the friend or relatives worked against the C.F. child e.g. a few mothers reported that grandparents were eager to look after the other children but unwilling to be left with responsibility for the C.F. child.

It was interesting that the influence of cystic fibrosis on the attitudes of these others, to the child, did not seem to depend on the child's age nor indeed, upon how ill he was. In the same context it was interesting that mothers who described slight modifications in these relationships were more likely to perceive them as presenting a problem, than were the mothers who described gross differences in the reactions of others to their C.F. children. It seems, then that family friends and relatives tend to follow the lead set by the attitude of the mother, in their own attitudes and behaviour to the C.F. child.

Family Communications (Section III.C.3.)

The point made at the beginning of the preceding discussion of family relationships is very relevant here too, for even before the advent of cystic fibrosis into the family, communication between family members, in some cases, was not good. It was unlikely to be improved by the influence of this anxiety-provoking disease. Poor communications between parents, then, described the situation where the fathers' movements tended to be independent of those of the rest of the family, i.e. tehy tended to take little part in the rearing of the children or in the activities in the home, and went out

frequently without their wives. These fathers then tended not to be present when the disease was diagnosed and their ignorance of the child's condition made communications between the parents possibly even more difficult than before. Poor communications with others, tended to represent strained relationships with others ouside the family, or to represent those who preferred 'to keep themselves to themselves' even before the children were diagnosed. Communication with the C.F. children, to some extent, depends on the degree of linguistic sophistication and intellectual maturity which they have achieved, as well as upon emotional or personality factors which might also hinder communication.

The interesting group from the point of view of this thesis, were those who communicated satisfactorily with others about other matters but experienced barriers of some kind, to aspects of their discussion of cystic fibrosis. Communication was best between parents but, even there, some difficulties could arise. Parents experienced little difficulty in discussing the practical aspects of the child's treatment but difficulties were described in discussing the implications of the disease, the child's condition or his prognosis. Mothers complained that their husbands would not discuss with them their worries about the future and fathers tended to avoid discussing their worries about the child, with their wives, for fear of distressing them. Turk (1964) and Kulcztcki et al (1969) distinguish parents whose communications were generally poor, from the other parents, but their findings were qualitatively, close akin to those reported here.

Turk however reported that the parents in her sample did not discuss either the child's condition or its treatment with other family members or with friends. Although several of the

parents in this sample were very conscious of not allowing cystic fibrosis to become an obsession, dominating their every conversation, almost half of them were able to discuss these matters with their families or friends. The difficulties, again, arose in discussing the child's future and parents sought to protect themselves from the curiosity of others by avoiding such discussions, in almost a third of the cases.

The difficulties in discussing the diagnosis with the children were more profound and only one fifth of the parents described no barriers to their discussions with their children. More than one third of the parents, who otherwise communicated well with their children, found cystic fibrosis a very sensitive topic. Their children had, naturally, asked a variety of questions about whether they would always need to have medicines, why they had to undergo treatment when their brothers and sisters did not and what was the matter with their chests or stomachs. These parents described considerable difficulty in replying to questions of this nature and some of them admitted resorting to evasive or deceitful tactics because they could not bear to tell the child the truth. e.g. one little girl has been led to believe that she has an ulcer, 'just like her uncles', and that she must do as the doctor says or it will not go away.

Clearly it is a problem, just what these children are to be told. Certainly unbridled candour can be extremely damaging and the child who is exposed to the possibility of a fatal prognosis is likely, naturally, to be very disturbed indeed. Nevertheless, it is not necessary to be brutal, in telling children the truth and we can learn a great deal from the parents who seemed to handle this situation honestly but gently. A simple

and truthful answer, given in an age-appropriate way seemed to suffice until the child was ready to ask for more information. Where parents described embarrassment or distress when the children asked questions, it was clear that such questions were unlikely to be repeated, and that the parents were relieved when the issue was dropped. Their children then, were described as, at best, only moderate communicators, again, taking their cue from the attitudes of their parents. However, there was evidence that the issue was not forgotten by children, as it could hardly expected to be, with treatment routines, clinic and hospital visits playing such a large part in their lives. If unbridled candour is disturbing so, too, is the 'web of science', for it denies the children the opportunity to be relieved of their worries. It is perhaps useful then to consider the approach taken by several of the mothers, among the good communicators to explaining cystic fibrosis to their children.

The essence of the explanation is that lots of people have something wrong with them. Some people have little things wrong e.g. "Daddy's eyes don't see very well so he needs to have spectacles to help him" and other people have more serious things wrong e.g. "that little boy can't walk/see/hear" (depending on child's experience.) In this context of the things that go wrong and what can be done to help, these mothers then found it easy to explain to the child that his tummy didn't work properly because there was something missing, so he couldn't digest his food. The child's experience of sore tummies or diarrhoea could also be incorporated in this explanation, which then was used to justify the pancreatic substitute and other medicines which the child had to take for this aspect of his problem. Lung problems

could be explained in an equally simplistic way, describing the need for physiotherapy in terms of the need to clear out the nasty thick mucus in the child's lungs to get rid of the germs.

Explanations at this level were clearly not necessarily all given at once, nor were they necessarily forced on the children who had not become curious, but as an example of a method of introducing the topic, they allow the mother to reassure the child that he is not so different from others, i.e. no-one is perfect, that something can be done to help him and that he is luckier than lots of people e.g. the little boy who is unable to walk/see/hear, because he can do all the things that other children can do. This last point may, of course, have to be modified in the light of any things which the child cannot do.

The importance of this issue really lies in preparing an open channel of communication wherein the child can discuss freely, any worries which may arise, over the years, in relation to his condition and can be assured of an answer which will help him with his problem. The basis for good communication in this matter depends to a considerable extent on the climate within the family when the topic of cystic fibrosis is raised and on the information which the parents themselves have about the disease and their own child's manifestations of it.

The whole area of information communication on the topic of cystic fibrosis is a delicate one which affects all who come into contact, or rather, into coversation, with cystic fibrosis children and their families. It is an area worthy of continuing study.

It is a constant hazard in studies of this kind, that the investigator may become so absorbed in the instances of behavioural pathology that he finds that he neglects to report them in proper perspective. We have already observed specific instances of the consequences of this event among research reports appearing in the literature and have endeavoured to guard against such a tendency in this report.

In terms of the impact which cystic fibrosis has on the lives of the affected families, it must be pointed out that a large proportion of families adapt to the child's needs and cope most admirably. Some of the 40% of fathers who described cystic fibrosis as intrusive in the lives of their families, were doing so on the objective grounds that their occupational decisions or geographical mobility had been influenced by consideration for the needs of that child. In the main, cystic fibrosis was felt to have an impact on family functioning that was not measurable in material terms, but which was felt in the somewhat sobering effect which the disease had had on the families' outlook on life in general. This was described by almost one in four of the parents. In practical terms, the brunt of the burden imposed is borne by parents, particularly by mothers and a lack of time for leisure activities was the next most commonly perceived hardship, with the personal financial sacrifices made by the parents for their C.F. child ranking third.

Where hardships or difficulties are occasioned for the individual family because of cystic fibrosis, whether in terms of financial stresses, social isolation, family relationships or the quality of life in general, then these problems should not be

ignored, for they can be real and important sources of stress to the family concerned, in a way which can only impede their ability to care for their C.F. child. In the broader view however, these problems should not be over-emphasised for the findings of this study suggest that the majority of families make the necessary adjustments to their mode of life quite successfully, and that the incidence and severity of their problems has not always been put in proper perspective in the research literature.

The Effects of Cystic Fibrosis on Parents (Section III.D.1.)

This study was principally concerned with two aspects of the effect of C.F. on parents, as parents. Parents' long term response to the diagnosis of cystic fibrosis, and the modifications which were thus introduced into their approach to rearing these children, were both of interest.

In the case of each of the children in the sample, the diagnosis of cystic fibrosis had been carefully confirmed to the satisfaction of the physician in charge of the case. Nevertheless, it was suspected that the lack of visible stigmata associated with many of these cases might lead to doubts, in those cases, about the validity of the diagnosis. Although the majority of parents said they accepted implicitly the doctors' diagnosis, almost one third of all the parents interviewed had experienced some very real doubts, occasioned in most cases, by their child's lack of obvious or troublesome symptoms. In several cases, this doubt was also acting as a defence against the harsh reality, as much as it was providing an objective comment on the apparent state of the child's health. In this context, parents sought to describe

how, at times, they felt that although they could understand the nature of cystic fibrosis they simply could not believe that this diagnosis really applied to their child. In most cases, these feelings were transient and could be regarded as a manifestation of the parents' wishful-thinking, or hope that their child could be cured. In a few cases however, where the denial was deeply rooted, such doubts could be troublesome. This was apparent in families which had two affected children of whom one was severely affected by cystic fibrosis and one only mildly so, and similarly, in the family where two severely affected children had died, leaving a third surviving child, also severely affected by cystic fibrosis. In each of these cases the doubts, based on the parents' experiences but also on their hopes and fears, could have deleterious effects on the standard of treatment which they felt that child's health required.

Parents' willingness to experiment with the child's treatment was not commonly found to be a direct expression of their doubt of the diagnosis. Fathers, on the whole, were quite happy to 'do as the doctor said', but several of the mothers commented that they had felt a desire to withdraw all therapy, just to see how well the child could manage without it. Although they often justified this desire with the motive that they were anxious about the long term use of the medicines, it was very rare indeed for medicines actually to be deliberately withheld. The situation was rather different in the case of physiotherapy, where several mothers remained unconvinced of its prophylactic value. Almost half of the mothers had allowed the child to go without this form of therapy for a time, at some stage of his life. In most cases, the therapy was resumed after a week or two, when the

child developed a cough or the mother's conscience began to prick her. Nevertheless, in some cases, this vital therapy had been withheld for longer periods without apparent detriment to the child's health. Certainly, the administration of physiotherapy can be tedious and time-consuming. If the child protests and the mother's efforts seem to be unproductive, it is not surprising that some of the mothers do begin to omit it. These findings suggested a need to remind mothers of the importance of pulmonary hygiene to the child's prognosis and, as a result, discussions of this nature were introduced at routine clinic visits, in an attempt to remedy this situation.

The parents would scarcely have been human had they not experienced periods of feeling over-burdened or discouraged by cystic fibrosis at some time. What was important was that they should also have an enduring underlying feeling of optimism or hope about the child's progress which would help to carry them through these black periods.

Tacitly, most of these parents, understandably, hope for a miracle cure which will save their child. This hope makes them very vulnerable to reports in the media which describe new advances in the treatment of cystic fibrosis and these announcements need to be very carefully worded, in order to avoid raising false hopes among C.F. parents. Doctors at the out-patient clinic in Edinburgh had had the unhappy task of enlightening parents who had been elated by the implications of such newspaper or television reports, on several occasions during the course of this study. This suggests that more caution is required in the issue of press releases about cystic fibrosis.

However undesirable unrealistic euphoria may be, the more moderate optimism expressed by these parents must surely be viewed as vital, for without hope, it is difficult to see how these parents could be expected to continue in their task of caring for the C.F. child.

Parents' attitudes to their C.F. Children (Section III.D.1.)

A key issue in studies of parent-child relationships in the case of children withchronic illnesses or disabilities has always involved the assessment of the extent to which the parents over-protected or pampered the child, because of his condition. (Section I.c.) We have already deplored the moral judgments implied by these assessments (Sdction I.E) and have endeavoured to describe the parents' views without imposing the values of the investigator.

Certainly there was a tendency among parents to hold different expectations of the C.F. child because of his cystic fibrosis, than they would have done had he not been so affected. This was perfectly reasonable where the child's health was poor and he was genuinely incapacitated by cystic fibrosis, though even in these cases it can be argued that many of these children are quite able to find their own limits without external sanctions. Nevertheless, it was clear that in many cases the parents' attitudes were not always modified in a way which was commensurate with the child's capabilities.

The tone of parents' attitudes to their sick child was initially set by the way in which the diagnosis was communicated to them and from the beginning most of the mothers described making

allowances for the child's infant behaviour e.g. feeding, toilet training, dressing or temperament, because they were so anxious about his health. If the child presented problems, as an infant, in any of these respects, they were likely to continue to make allowances for them. Some of the specific questions referred to challenges which faced only the older children, but parents of pre-school children described what they hoped would be their attitudes to these issues and, in so doing, showed no significant differences from the expectations held by parents whose children had reached these hurdles.

Parents tended to place less emphasis on the importance of school achievement for these children. Although it was only natural that they should give the child's health first priority, it did seem that this attitude was the product of a rather blinkered view of the future. Since these children are better able to make their way in the world by their brains rather than by brawn, it would seem that educational values should still be relevant. The child's abilities have to be considered too, though, and the two parents who replied that, on the contrary, they wanted their C.F. child to do well at school to compensate for his physical weakness, were courting disappointment.

Several parents expected considerably less of their C.F. children in terms of their activities outside the home. They were less likely to encourage these children to join peer group activities, especially if they involved physical exertion and, if they took place away from home. Since these attitudes were more likely to be discouraging if the child's physical condition was poor, it might have been thought that the parents' views were amply justified.

However, within the experience of this study, there was a problem of morale, especially among the older children in poor physical condition. This problem was not helped and indeed was aggravated, if the children were encouraged to be inactive and solitary spectators in life.

Parents who had encouraged the children to be active and sociable, sometimes even against the children's own lethargic tendencies, were pleased to report that their peers were quite happy to accept the children and to allow them to find their own physical limitations.

The question of the punishment of C.F. children was a vexed one. From the beginning, mothers said that they were afraid to scold or punish their C.F. infants for fear of exacerbating their symptoms. Several of these mothers later came to regret their laxity, as they reported that the children could learn to play on it, while the healthy siblings could become resentful. Even among the parents of older children there were those who claimed to be pleased that their C.F. children were well enough to be naughty. There was then an air of uncertainty and inconsistency about this issue throughout all the families in the sample, and a high proportion of mothers expressed a degree of dissatisfaction with the way in which they were bringing up their C.F. children. This uncertainty seemed to be, initially, the product of the complete lack of confidence experienced by mothers in the early years, in the area of discipline and it seemed this lack of confidence was never quite resolved. Although a number of mothers claimed that their C.F. children rarely needed to be disciplined it was fairly clear that they would be uncertain how to handle any problem behaviour which

did require correction. It was hoped that some of the anxieties about these aspects of child-rearing policy might be settled by the generally reassuring effect of the new home-visiting service. In this respect, it was hoped that this service would not take over the responsibility but, rather, that it would give these mothers the confidence to carry out in practice what they thought to be right in principle, in a way which some of them had been hitherto unable to do.

Fathers' participation (Section III.D.1.)

Not all fathers were involved in the care of their C.F. children. Perhaps this is not surprising, since so many of them were not able to be present at the time of the child's diagnosis, nor, commonly, at his routine clinic visits. Certainly it was the minority of fathers who had learned how to administer physiotherapy, although those who did tended to take a fair share of the load. Several fathers did express a willingness to participate more in the child's treatment if only to relieve their wives on occasion, and it seemed that they required only a little tuition and encouragement to enable them to do so.

On the whole, the fathers in this sample seemed to be willing to assist in other domestic tasks though usually not on a regular basis and there was still a sizeable proportion of fathers who took little part in the rearing of the children or whose contribution was only to take the children out in order to free the mother for other tasks.

Clearly, in our society, the traditional roles of mothers and fathers, husbands and wives, are changing. Under the additional pressures created by the need to provide treatment for a child with

cystic fibrosis these changes bring welcome support to mothers on whom so much responsibility rests in the care of these children.

Parents' Groups (Section III.D.1.)

The Cystic Fibrosis Research Trust is primarily a fund-raising organisation which seeks to sponsor research which will produce results of benefit to cystic fibrosis children and their families. Regional Committees of this Trust do organise meetings for parents and supporters living in the same geographical area and when we refer to the parents' group it is to these meetings that we refer. At the time of the study these meetings occurred at irregular intervals and were not well supported, except by a small group of parents whose children had been diagnosed some time previously. The members were uncertain of the function of their group and they were anxious both to clarify its role and to attract new members. Some of the findings of this study, although, perhaps, already out of date in the light of developments which may have occurred since the study was completed, may be enlightening in this respect.

In the first instance the C.F. Trust was not well known among these families. Families whose children were patients at hospitals other than the Royal Hospital for Sick Children were unlikely to know of the Trust's existence, and even among those attending the C.F. Clinic at that hospital, a large proportion had not been aware of the parents' meetings. If the Parents' Group was to be a viable proposition it was clear that its meetings required better advance publicity.

However, there were some obstacles to parents attending meetings, even when they were informed of them. Parents described practical difficulties which barred them from attending the evening meetings which, at the time of the study, were held in a lecture theatre in the hospital. Curiously enough these factors did not significantly distinguish members from non-members, although admittedly it was harder for families who lived further away, or who had large families to attend to, to come into the hospital for an evening meeting. Nevertheless, it was discernible that practical factors were not the only barriers. Another problem was the social class composition of the group. As it had evolved, representatives of social classes I and II predominated and the wives of manual workers who had, on occasion, attended meetings of the group explained that they had so little in common with these mothers that they had not returned to subsequent meetings.

Before these practical problems could be tackled it was important, though, to establish just what the aims and functions of these meetings were to be, and to what extent parents' supported the basic concept of a Parents' Group.

Social psychology suggests that under anxiety-provoking conditions people show affiliative tendencies and, moreover, that these tendencies are directional. People who are anxious are likely to show a desire to be with people who are in the same situation. (Schachter, 1961) Schachter has delineated five reasons why this should be so, which may help to clarify our thinking about the functions of this parents' group. He labels these escape, cognitive clarity, direct and indirect anxiety reduction and self-evaluation. The first, escape, is not relevant to the situation in hand, since the group members cannot opt out of being parents of children with

cystic fibrosis. The members may join the group for information (cognitive clarity), social reassurance (direct-anxiety reduction), social diversion (indirect anxiety reduction) or to help them to come to terms with cystic fibrosis (self-evaluation.)

Although it has already been established that mothers, particularly, did feel some, if not all, of these needs, there were doubts about whether the Parents' Group was the best means by which they could be met. In terms of the social functions, one of the problems was mentioned previously, in connection with the children's hospitalisation. Several mothers found the group meetings depressing: if another mother's child was ill, it acted as a reminder the problems that their own child might have to face; if the other child was doing well, then, the mother's own child seemed to be a poor case in comparison. Some mothers had not, so far, had this experience and they did find it helpful. There was a strong relationship between mothers' attitudes to meeting other mothers and their attitudes to the concept of a parents' group, and although, the meetings at the time of the enquiry seemed to be fulfilling a primarily informative function, it was these social attitudes to meeting like others which sharply distinguished members from non-members. To this extent then, the principles of social psychology hold good.

Although there had been a considerable importance attached to the informative function of these meetings, it was clear from the discussions which followed these meetings that parents did not really want general information about all the things that could present complications in cases of cystic fibrosis, they wanted to know about their own children and their prospects. There was a danger here too,

that the informative aspects of the meetings were of greater benefit to the more intelligent, more articulate parents, usually of social classes I and II who tended to dominate the discussions at these meetings, while the other parents, who had equally as many problems in relation to C.F., left the meeting, as they had arrived, afraid to ask questions about things which troubled them for fear of sounding stupid.

Of the fund-raising activities parents were divided. Some mothers found these helpful, in that the proceeds allowed them to feel that they were making a valuable contribution towards helping children with their child's disease, and indeed they were. Individuals and small groups os parents enlisted the help of others, and were very effective in this aspect of the group's activities, but, there was rather a lack of enthusiasm for fund-raising among those parents who were enthusiastic about the social and educational functions of parents' meetings. Clearly a division existed. It was, in a way, unfortunate that only the views of mothers had been gathered about the Parents' Group, since a sizeable proportion of those who attended, came with their husbands. Fathers tended to be most vociferous at the meetings and fathers had always taken the Chair.

This was a rather different situation from those meetings described in the literature e.g. by Linder (1970) where the groups consisted entirely of women. Successful parent meetings, of mothers of children suffering from conditions of comparable severity to C.F., have had many of the qualities of therapeutic groups, and while they have been notably successful in American society, it is doubtful whether they would be acceptable in the terms of the cultural and social milieu of this sample.

The group discussions described by McCollum et al (1970) seemed less traumatic to parents and possibly more acceptable to the families in the sample. At these sessions, the emphasis was still on the exchange of views between parents, as in Linder's group, rather than on the provision of an authoritative source of professional information, and members of the group introduced topics into the discussion quite spontaneously. Although shared attitudes and feelings aroused by cystic fibrosis were discussed, the discussion was attended by a doctor or by psychiatric social worker, from the team who cared for the children, who imposed limitations on the depths of feelings explored. It was quite reasonably said that "the exposure of the deep fears of one can threaten the defences of the others". This then seems to be only a tentative attempt at an encounter group, and there would still seem to be some obstacles to the introduction of such a system in Edinburgh.

The picture which emerges in this study suggests that the needs for information and social and emotional support, at least of mothers, may well prove to be met by the new system of home-visiting, just described. Information about research developments may equally well be disseminated by written report and by individual discussions with the doctors, as by public meetings, and it would seem, that the concensus of parent opinion at present, suggests that there is less in favour of continuing these meetings, than there is against them. Fund-raising activities, requiring larger group organisation than the present-scale of activities, are another matter, not adequately covered by the scope of this study.

This suggestion that the Parents' Group Meetings have outlived their usefulness, is based on mothers' reports. It was

clear that mothers' needs for discussions at the domestic level of management were never met by these meetings in any case, and that the mothers were frequently emotionally upset by these evenings' events. This excludes the needs of fathers. It has been clear from the findings so far that there is a need to encourage fathers to participate more in the care of their C.F. children and, to that extent, discontinuation of these meetings would remove an opportunity for them to play their part. Nevertheless, from the distress caused to mothers, at no obvious gain, by these meetings, the conclusion, for the moment must remain, that there is evidence to suggest that Parents' Group Meetings fail to meet any of the major needs of parents of cystic fibrosis parents.

This conclusion could be interestingly reviewed when the home-visiting service has become established, in order to determine whether there are any outstanding needs, beyond the desire to raise funds, which are still then unsatisfied.

One point which was made by several mothers has been ignored to date. This was the converse of an earlier-mentioned difficulty, hat the mothers were eager to see children who had cystic fibrosis, who were older than their own child and who were relatively well and happy. Although the C.F. Trust's Newsletter endeavours to meet this need, it tends to describe the activities of predominantly English children. The mothers seemed to require further reassurance that children in their own region could also do well. This need was apparent in the record attendance of parents at a group meeting at which it was announced a young woman with cystic fibrosis would attend. This young woman was married and had a normal child, and clearly she epitomised the hopes of all the

mothers of C.F. girls who attended that meeting. Some further thought needs to be given to ways in which this great encouragement can be given to parents from time to time.

Cystic Fibrosis and Parents' Marital Relationship (Section III.D.2.)

The genetic implications of cystic fibrosis have already been discussed and it was clear from other research conclusions, that there was a need for caution in this area of the study. It is simply not possible for the outside investigator to assess the happiness of a marriage anymore than it is possible for him to identify which of a myriad of factors has been primarily responsible for marital breakdown. Although cystic fibrosis as an abnormal element may be pinpointed as being responsible for the marital difficulties, even by the parents themselves, the findings of this study would suggest that the difficulties, where they exist, are usually multifactorial. Since an antidote was clearly required to the unmitigated gloom of these earlier reports, this study ventured a little way into the exploration of the effects of the child's illness on the parents' marital relationship.

Some tensions were created over the problems of communication already mentioned and these could be quite distressing. Although difficulties sometimes arose over the extent of fathers' participation in the children's upbringing these did not seem to have a particularly disruptive influence. About half of the parents rarely went out together for pleasure but it really did not seem that cystic fibrosis had seriously curtailed parents' private activities. A few mothers did say they had no-one whom they could trust to care for the child in their absence and some parents cooperated to let each other out separately, although they rarely

seemed to go out together. The conclusion was therefore that parents who wanted to go out in the evenings for their own leisure, were usually able to arrange to do so.

Although some mothers did describe a lack of energy and a persistent anxiety lest the child require comfort during the night, in a way which interfered with the physical aspect of their relationship with their husbands, the most worrying issues in this respect were those which concerned the mothers' fears of further pregnancy.

On the whole then, only 29% of the mothers felt that cystic fibrosis had intruded in their marital relationship. The proportion of men who felt this to be the case was rather higher (43%), possibly because they saw more of the effects of stress on their wives than these women themselves appreciated. The findings certainly suggested that cystic fibrosis could be an important contributory factor to the breakdown of marital relationships which were already strained, but it was never the sole factor, a point often omitted by previous researches (e.g. Lawler et al, 1966.) Indeed it was encouraging that, given the opportunity to say so, 42% of mothers and 34% of fathers felt their marital relationship to be stronger for the responsibilities they had shared.

The Parent as an Individual (Section III.D.3.)

Although we have observed that in some respects, being able to participate in the child's treatment has a therapeutic, anxiety-dissipating effect on parents, the tremendous personal and physical involvement required can pose a considerable burden, particularly on mothers. If parents are trying to meet all their

other responsibilities to the full as well, then one can surmise that the personal toll paid will be high. The findings of this study support this view.

Certainly in the immediately post-diagnostic period the parents reported disturbances in their appetite and sleep, during the period of anticipatory mourning. However, even as the family settles to its pattern of long-term adaptation, mothers, in particular, described recurrent periods of depression associated with cystic fibrosis, and an underlying more permanent change in the state of their 'nervous activity.' These terms were used in a lay sense, but in many cases the troubles which they described had been sufficiently severe to prompt the mothers to seek medical advice. The relationships linking these problems, the child's health and the mother's level of anxiety seemed fairly clear, even from the mothers' own accounts and they were borne out by the analysis of the findings. The feelings described by mothers, of a general increase in emotional ability, from the time of the diagnosis, followed thenceforth by periodic depression or 'nervous tension' were indeed those of common experience, documented by other research workers (Rosenstein, 1970; McCollum and Gibson, 1970.) These periods of particular difficulty were sometimes triggered by outside events e.g. a reminder of the possible prognosis but it seemed that they could also occur for no consciously apparent reason. Fathers were much less likely to be troubled in this way, and it was likely that their ability to tolerate the symptoms of stress shown by their wives was an important facet of the strength shown in their relationship in the face of cystic fibrosis.

Three aspects of coping behaviour had thus been considered,

in the issues of practical care, family responsibilities and personal integration. Although it was clear that some mothers were able to cope admirably, functioning well at all three levels, many mothers did have some difficulties. The children were, on the whole, quite well cared for and the most common problems stemmed from the interplay of the factors concerned in mothers other responsibilities and in their state of mental health.

Although there are no obvious answers to the problem of how the stresses on these mothers may be alleviated, this study has taken the approach that by seeking to provide relief in the public and practical aspects of the problem, we may hope to release additional reserves of strength in these parents, to enable them to meet, themselves, the more personal and emotional problems which they face, in coping with cystic fibrosis. If then the emotional problems prove to be invincible, then it will be necessary to review once more, the parents needs with a view to providing further specialised sources of help.

The Influence of Cystic Fibrosis on Infant Development (Section III.E.1.)

Young children with cystic fibrosis can, quite objectively be difficult to rear. Although the retrospective method of assessing the nature and frequency of these difficulties was not entirely satisfactory, there was little reason to suppose that mothers would say that they had problems when they did not, so that the findings of the study should present at least, a brief resume of the likely areas of difficulty.

It was clear that several of the mothers were anxious about

the 'finicky' eating behaviour of their children at this stage, which was much more commonly described in this sample than among normal children (2-5 year olds, Illingworth, 1972; 4 year olds, Newson and Newson, 1968.) Although, in such a heterogeneous age group of children, it was difficult to define toilet training problems objectively, mothers were usually quite clear about when this aspect of their C.F. child's behaviour did not reach their expectations for that of a normal child of his age; and this was counted as a problem to the mother. The deviant nature of the children's stools and the tendencies of the children to frequency and urgency made it difficult for mothers to assess the extent to which the problems were symptomatic of cystic fibrosis and to which they were indicative of resistance to training on the part of the child.

In this context, it is interesting to note that Illingworth (1972) has recorded that between 10-20% of normal children of this age range, present just such feeding and toilet training problems and he has identified parental attitude as an important causative factor. Over-anxious, over-protective and indulgent mothers as well as those preoccupied with the child's growth curve or with the nature of his stools are, he records, more likely to bring these problems on themselves. Certainly, the mother of the C.F. infant has had good reason to be all of these things. Difficulties in obtaining a diagnosis, and the schock of that diagnosis when it is obtained, leave her distressed and anxious. Fear of losing her child makes her both indulgent and protective, while her part in his treatment necessitates a greater involvement in his weight gains and bowel habits.

The findings of the Newsons (1968) would tend to support particularly this latter point and it is hoped that, among her many other functions, the home-visiting nurse will be able to take the high emotional charge out of some of these traditional areas of difficulty. Although it seems likely that the disease presents some real physical basis for these problems, in many cases it is also clear that the mothers have a particularly high personal investment in the development of their children, at this stage which can act to exacerbate existing problems.

Of the repertoire of devices that children of this age use to capture attention, Illingworth (1962) suggests that these tricks indicate that the child is not being recognised sufficiently as a person, nor being given the responsibility he wants. This study did not provide a sufficiently detailed account of the development of the children to be able to suggest any causal relationships, but it was interesting that the mothers, particularly of slightly older children, did tend to report that they had done more for their C.F. infants than had been really necessary.

Although these views were reiterated when the children's social development was being assessed, their scores on the Vineland Social Maturity Scale did not show any statistically significant deficits. However, the sample of children under the age of five years, at the time of the study, was small and the validity of the norms was questionable, so that more information was gained from the mothers' views than from the numerical analysis.

Even at this young age there were indications that the development of social behaviour could be delayed. Mothers were naturally anxious about the risks of exposing the child to other

children's germs and the children themselves seemed to tend to timidity, so that the combined effect was a reduction in the social experience of many of these infants. With hindsight, several of the mothers of older children felt that they had been over-cautious and over-protective in their handling of their C.F. infants and it is this aspect of the influence of cystic fibrosis on infant development which we should seek to alleviate.

C.F. Children and their Illness (Section III.E.2.)

The mothers of all of the C.F. children in the sample described a range of symptoms of the disease which they had found troublesome during the course of the children's lives. However, from the point of view of the older children who were interviewed, there was little doubt about which symptom was most troublesome. The odours created, both by flatulence and the children's stools, were a frequent source of great embarrassment to the children. Although some of the older children were able to comment on these difficulties, the younger children simply nodded rather miserably when they were asked about toilet problems. The children who had persistent and productive coughs found this symptom also to be considered socially unaccetable by strangers whom they met in some spheres of their lives but, the problem did not seem to be so common nor, indeed, so shameful to them, as the toilet problems.

The children who were interviewed had all already begun to function independently outside their own homes, if only in the school environment, and it was in this context that the need for medication and for regular clinic visits began to appear strange to them, in the light of the activities of other children. It was embarrassing, then, for the children, in many cases, to have to

take pills to school and where it was left to their discretion, they would either omit to take them or seek the privacy of a corner to swallow them unobserved. The time off school for clinic visits, another distinguishing feature, was a further source of embarrassment since it aroused the curiosity of the children's peers. The majority of the children were defensive about these excursions and tended to tell other children to 'mind their own business' if they asked questions. Older children were quite glad of a special friend or group of friends in whom they could confide, but they too said that they evaded the questions of more casual enquiriers. The children tended to follow their mothers' lead in this however, and where mothers had stressed that there was nothing shameful about having cystic fibrosis then the children seemed more likely to have the confidence to be open about it, too.

The point remains however that both the symptoms and the treatment of cystic fibrosis can be acutely embarrassing to these children.

By the time of going to school most of the children seemed to realise that there was 'something wrong' with them. How soon this notion developed depended on several factors e.g. the severity of the child's condition, his experience of normal children and the attitude of his mother, but unless there was a deliberate policy, made by the parents, to keep the child in ignorance, the children then developed age-appropriate interpretations of what, indeed was wrong. By questions and by observation, then, they built up a picture of cystic fibrosis, although the level of understanding achieved by the time of the enquiry did not seem to be so good as

that reported by Tropauer et al (1970), where 83% of the sample were said to show a good appreciation of C.F. for their years. Here, a substantial proportion (56%) of children over the age of five years, still had very little understanding at all.

Since the literature had so stressed preoccupations with death among C.F. children e.g. Lawler et al, 1966; Kulczycki, 1969, a key issue was then to determine the extent of the children's awareness of the possibility of a total outcome to their condition. Clearly this was not a piece of information which parents were likely to volunteer. Nevertheless six of the mothers knew that their children were aware of the prognosis, three of them having been rather tactlessly so informed by a thoughtless sibling or by someone outside the family. Two other children also indicated to the investigator that they were aware of the possible outcome of cystic fibrosis although they had clearly not communicated this knowledge to their mothers.

The question of how children respond, both to the symptoms of cystic fibrosis and to their knowledge of the disease, was an interesting one. Although mothers had described most of the preschool children as being rather pleased to find themselves the focus of attention, among the children interviewed the investigator found only two who seemed to delight in the attention that C.F. brought to them. The more usual reaction, as the children's horizons broadened outwith their homes, was for them to resent the intrusions which cystic fibrosis and its treatment made into their lives. Self-consciousness included not only the aforementioned problems in relation to symptoms and their treatment, but it also developed in relation to the child's physique. Thin children, small

children and those with distended abdomens all felt self-conscious about their bodies and later, this anxiety about self increased if secual development seemed to be delayed.

The most marked reactions seemed to have been shown by the older children at some stage during their lives, and not surprisingly, the most dramatic responses were occasioned by the exposure of the child to the life-threatening nature of the disease. This knowledge seemed to mark a turning point for the children and they were indeed given to periods of black melancholy at times thereafter, causing their parents considerable distress and anxiety. Not even these children however showed the enduring preoccupation with death described in the literature.

As in the communication of the diagnosis to parents, so to children, the conduct of this interpersonal event is highly dependent on the personalities involved and no firm rules can be given. Nevertheless, the findings of this study suggest a need for children to have good communications with their parents in such a way that the child's worries can be discussed and alleviated and his trust in those who care for him may be encouraged but, in such a way that feelings of shame about having cystic fibrosis may be dispelled. Although candour would then seem to be indicated a caveat should be taken from the devastating effect which learning of the prognosis has had on some of the children in this study. If communications are good, it may be hoped that this issue too could be dealt with, should the need become apparent, but this study offers little justification for offering such information to children.

The nature and extent of the knowledge with these children have about cystic fibrosis can be seen to be an influential factor

in the subsequent course of their personal development.

The Education of Older Children (Section III.E.3.)

In the first instance, this study confirmed that the intellectual capabilities of children suffering from cystic fibrosis show a normal distribution of scores when assessed by a standardised psychometric test.

There was no evidence to support the thesis of Spock and Stedman (1966) that the children are particularly verbal. On the contrary, the children tended, rather, to obtain high performance scores, which seemed to mirror their interests in creative and constructive indoor pastimes.

The children were tested during periods when they were well i.e. attending school, in order to optimise performance levels. They were, without exception, able to relate quite satisfactorily to the investigator, to maintain their concentration throughout the tasks set them (unless interrupted by outside events) and to complete the test battery as it was presented to them, without signs of fatigue. Most of the children seemed highly motivated and willing and indeed several of them said that they had enjoyed the experience. In the home setting then there was very little evidence of restlessness or distractibility among the children tested.

In tests of school subjects the childrens' performance was less satisfactory. However, a few of the children had begun to learn to read on the i.t.a. (initial Teaching Alphabet) and, having only recently made the transition to the common alphabet, they found Schonell's Reading and Spelling tests

particularly difficult. As anticipated, several of the older children found some of the measurements, used in sums in Vernon's Graded Arithmetic Mathematics test, rather unfamiliar. In these cases, lowered scores were to be expected and it was for these reasons that it was decided to recognise underfunctioning only when the children were behind the norms for their age and intelligence by a year or more.

Several factors were identified which seemed to contribute to this depression of the childrens school performance. Certainly illness would be expected to interfere with schooling but children who had had longer spells in hospital seemed to fall behind only in the acquisition of numerical skills. Their reading and spelling scores were no more likely to be poor than were those of the other children, and when clinical condition, as indexed was used as an assessment of illness then no significant relationship was established between the general standard of the child's health and poor school performance in any of the subjects tested. This may in fact be an oversimplification since some of the mothers of children in poor physical condition did explain that their children did seem to have 'off-days', when they were very difficult to stimulate and would probably perform badly at school.

There was a strong suggestion that the more intelligent children seemed more likely to perform badly. This was rather curious until one recalls that emotional factors can, commonly, interfere with children's school work. Within the limited span of this study it may be suggested that the more intelligent children, with more enquiring minds, are more likely to ponder

on cystic fibrosis and perhaps, therefore, are more likely to show emotional reactions to their condition which may interfere with their school work. The evidence for this suggestion is largely based on observation and it requires to be submitted to more intensive examination in a larger sample.

A further point in the children's school performance may be made as a reminder of the factor of parental attitudes. Certainly there is a trend for these parents to de-emphasise the importance of school achievement for cystic fibrosis children so it is not surprising that the incidence of parental permissiveness correlates fairly highly with poor performance. Certainly, from the observations of this study, the children did not seem to lack motivation, although the ability to sustain this motivation could not fairly be assessed; they may however lack the parental encouragement which would enable them to see school work as a challenge to be met.

Although on the whole the children liked school, it did present some problems to some of them because of cystic fibrosis. It did seem that where the parents and teachers were aware of the child's difficulties, they could cooperate to minimise them, but nevertheless, unhappiness at school could be another factor in the children's poor school work. Should this prove to be the case it is encouraging to find that this is a factor more amenable to remedial treatment than some of the others.

Teachers were not formally interviewed during the course of the study but the investigator took every opportunity that was offered, to visit schools to deliver the Bristol Social Adjustment

Guides to the teachers in person and it was possible to discuss their views informally on these occasions. Several of these teachers had been given minimal information by the parents about the nature of the child's disease or of its implications for the child at school. Others had, it seemed, been given a bewildering and anxiety-provoking battery of instructions for all eventualities and, on the whole, the teachers met, representing just over half of the school age sample, were rather more anxious about their responsibilities than perhaps the mothers realised. Only a few teachers insisted that the child was treated exactly as a normal child, as the mothers had indicated, and more of them were inclined to relax their demands of the children if they felt they were in any way "off colour."

This permissive attitude of teachers may also diminish the children's incentive to do well in school work, and may explain why several of those who performed poorly on the tests for this study, were described by their teachers as producing 'good' work.

This question of the reasons for poor school work can really be subsumed under the much wider moral issue of whether parents should take an essentially hedonistic approach to the upbringing of their potentially, dying children. Ultimately, this is a decision which only parents can take. However some of the findings of this study may suggest to them that, in the light of the ever improving prognosis for C.F. children, this is too short-sighted a philosophy which may ultimately defeat its own aims. Some of the unhappy results of well intentioned molly-coddling may be identified in the poor social and emotional development made by some of the older children.

The Social and Emotional Development of Older C.F. Children.

Cystic fibrosis seems at this stage, to constitute more of a problem to boys than to girls, perhaps because physical inadequacy contravenes the norms of social and personal development for boys more than it does for girls. Certainly among the children in this sample who were over 5:0 years old at the time of the study, it was the boys who presented the higher incidence of problems of this nature.

It was interesting that the early trends to poor social development seemed to continue into the older age group. These children were often described by their mothers as 'loners', playing at home and alone, at an age when, for normal children, the peer group should be assuming increasing importance.

Many parents admitted being protective about their C.F. children and hence allowing them less freedom of activity but it was clear that often the children themselves were timid about the rough-and-tumble of normal children, and were unable to stand up for themselves against physical, or even verbal, assault. It seemed that the children, especially the boys, were thus made more aware of the physical limitations which distinguished them from their peers.

Withdrawal from peer group activities encourages, as a corollary, interests in solitary, usually sedentary indoor pastimes. Although many of the children were quite happy reading or using their hands in creative and constructive play, these activities do not benefit their health. Parents were generally very cautious about allowing their children to take part in anything which would cause them physical exertion even though

active participation in games and sports is encouraged by the clinic. Although 47% of the boys were said to play football in several cases this meant kicking a ball around occasionally with a brother or a few friends, not sustaining a full team game. Girls in the sample on the other hand, tended to be more active in sports, joining in skating, ski-ing, netball, hockey, trampolining and even athletics. For those who did take some active part in sport, mothers generally reported some benefit to the child's health, even where this was graded C on the scale, and, at least for the girls, the sport served the additional function of breaking their social isolation.

Since the parents' attitudes and sanctions clearly delineated the opportunities open to the child at home, it was interesting to learn how these children then responded to the school setting, where they had to fend for themselves.

At school C.F. children tend to be described as showing under-reactive patterns of behaviour. Both boys and girls obtained higher scores in this mode of maladjustment than either their matched controls or their Stott's norms (1971.) This tendency was more marked among boys in the sample. These boys then tended to be more unforthcoming, withdrawn and depressed to a greater degree than either their controls or C.F. girls. This under-reactive behaviour was most marked among boys in poor health and was exemplified by such unusual behaviour as the tendencies to wander off alone or to be too timid to be any trouble.

The C.F. boys showed no significant differences from their controls in terms of over-reactive behaviour although both groups scored more highly than Stott's norms. C.F. girls on the

other hand, tended to obtain higher scores than either their controls or Stott's norms. The core syndrome most frequently scored by all the C.F. children in this category was that of Inconsequence. Stott explains Inconsequence briefly in the following terms, as:

> "A failure to inhibit first impulses for long enough for their consequences to be foreseen. The child seeks unthinkingly to gain attention, to dominate over his age-peers and to create an impression by showing off. In his school work he is apt to guess rather than to take time to work out thoughtful solutions." (Stott, 1971)

Since the numbers of children involved in these assessments was small no sophisiticated statistical analysis of the scores was possible. By observation, it seemed that the younger boys and those in better health seemed more likely to show this pattern of Inconsequential behaviour while older boys and those in poorer health were more likely to be withdrawn and depressed. One may surmise that the child's knowledge of his disease and its prognosis may be a contributory factor in this change. Girls on the other hand seemed when they were young, to be described as timid, quiet and unforthcoming while it was the older girls who were more likely to show inconsequential behaviour. Among girls the relationship between behaviour and clinical condition was less clear. Only further research will be able to confirm whether this is a truly developmental trend, differentiating the sexes in their response to cystic fibrosis, or whether as may be, this finding is an artefact of the particular personalities under investigation. Happily, few

of the children showed sufficient disturbance in their behaviour to warrant psychiatric referral, but there was sufficient indication of maladjustment to justify more attention being given in future to this aspect of the children's development. Behaviour disturbances of the nature of those described may also be contributory factors in poor school performance.

It was important then to examine those precursors of good and poor adjustment, which were amenable to change, in a way which would benefit the children. Certainly, some of the practical issues of having cystic fibrosis were so acutely embarrassing to the children that they were unwilling to discuss them even with their parents. The sensitive issues of having to take medicines, to remove clothes for gym or swimming or of having to use school toilets could all contribute to the children's unhappiness at school. However we have already noted that where they are aware of the problems parents and teachers can cooperate to minimise them. The more fundamental question is that of levels of communication.

Children whose communication with their parents was poor were more likely to be under-reactive and highly anxious. Experiences during hospitalisation, or the sudden exposure of the child to the possibility of a fatal prognosis could most easily evoke this withdrawn and anxious syndrome, especially among older children, and at these times good communications were found to be particularly important.

Dr. Burton has observed that preoccupations with death were more pervasive and emotionally more widespread among her Irish sample. It is interesting to note that experiences of

hospitalisation were more frequent and deaths more common among these families yet communications were poorer than in Scotland, suggesting that this point is indeed a valid one.

Even in Scotland though, a snumber of mothers were concerned about their children's emotional development and the children themselves did reveal some fears and worries about their condition. The incidence of hospital-related fears was understandably high and the majority of these children described fears of being ill or of having to be admitted to hospital. Many of them mentioned their anxiety even at routine clinic visits, lest they be referred to the hospital ward for admission, thereby vindicating the point made previously, about the bias that is introduced by testing children in the clinic environment.

The children's drawings, fantasies, dreams and wishes revealed very few of the abnormalities described in the literature. It was interesting though, that one boy of eight years, who was in poor health and presenting social and emotional problems, did show disturbances here. The child's drawing of himself was rather sinister, he described rather sinister dreams of witches coming to take him away to their home in the sky, and he would not answer any questions about his condition, his hospital experiences or even about what he would like to be when he had grown up. On the basis of this study then we cannot deny that emotional disturbance among C.F. children may occur and may be tapped by these projective techniques but in the course of this study the children's responses, where they were offered, tended to be mundane.

Given three wishes the older children all expressed a desire to be free of cystic fibrosis and they did worry about what was going to happen to them in the future but such responses were restricted to relevant questions and did not seem to represent the dominant theme in their mental processes, either at the conscious or sub conscious levels.

Emotional disturbances and morbid preoccupations do not, then, characterise this sample of C.F. children. Nevertheless there is no room for complacency, since a number of the children do show more moderate trends in the same direction. The children most at risk are clearly the older, the more intelligent and the more ill children who are more likely to become aware of their prospects and react accordingly. The findings already describing the early development of C.F. children may now be linked to those describing the oldest children in the sample to suggest means by which the children could be, in future, more effectively armoured against such disturbances.

A Note on the Problems of C.F. Adolescents.

A major problem seems to lie in the fact that so many parents (65%) regard making future plans for a C.F. child as merely tempting Providence and some are actually distressed when the children mention their hopes for the future. It is not surprising then to find that the two young people who had left school by the time of the enquiry were unemployed and that the other older children were anxious about their job prospects. Clearly vocational guidance should be made available to these young people, but to be really effective in constructing a

future for C.F. children we need to begin much earlier, by encouraging educational development to the full extent of the children's mental and physical capabilities. Then, by encouraging a more forward looking attitude among their parents, vocational guidance could really help the children to develop their talents in appropriate directions.

Social isolation and emotional unsettledness have been described among these children during the early years of school. In considering the prospects for adolescents we are reminded of the effects of stunted physical growth, delayed sexual development and prolonged dependence on parents at a time when contemporaries are growing up and gaining their freedom. In common with the adolescents described by Pinkerton (1969), Patterson (1969) and Teicher (1969), the young people in this sample were somewhat resentful of their lot. They chaffed at the protective restrictions imposed on them by their parents and were anxious about how they would ever achieve autonomy when treatment enforced dependence on others. Loneliness, anger, hopelessness, helplessness and depression were much greater risks to this age group. The oldest girl in the sample had renounced all treatment at the time of the enquiry and another two of the older children had had periods of refusal to cooperate in the past but had been since won around. The situation of these young people was not helped by the restrictions imposed from outside. Even where parents were willing to consent, schools seemed unwilling to take the responsibility of including C.F. children in their school outings or holidays, particularly if these involved foreign travel. Even where the school could be persuaded to take the child,

parents reported difficulties in obtaining the necessary insurance cover for the child. Although one of the children had been away from home for a week with a school excursion difficulties of one sort or another seemed to have barred most of the other children from such treats.

We would submit that as for educational and vocational problems, these social and emotional difficulties are better tackled much earlier than at this stage. The young C.F. child needs to be encouraged to join his peers in social and recreational pursuits and to enter friendships wherein he can be accepted for himself. Then, perhaps he would be better equipped to tackle the hurdles posed for him in adolescence.

Although many families cope admirably and do try to encourage independent activity and self responsibility in their C.F. children and although the picture posed by these children is indeed more hopeful than that described by some of the previous authors, there was a need to highlight the risks which cystic fibrosis, both directly and indirectly, can extend to the development of these children. These problems suggest little reason why parents and teachers cannot now be helped to improve the personal and social outlook for C.F. children by as much as the medical profession has improved their clinical prognosis.

The Healthy Siblings (Section III.F.)

In families where one child was affected by cystic fibrosis and where one or more other children were not, mothers frequently described having felt, at some stage that they had neglected their other children in some sense, in order to care

for the needs of the sick child. The times when these feelings were most common were when the C.F. child was very young and subsequently, when he was ill. Even where mothers did not feel that their healthy children had been left the rather special situation of these children was deemed worthy of some attention. In this study two aspects of their reactions were considered; their behaviour towards their C.F. sibling and their reactions as individuals in their own right.

In sibling relationships, it was clear that ordinal position was an important factor. Younger siblings predominantly tended to be jealous of attentions shown to their elder brother or sister. In most families in the study they were rather too young to understand the reasons for them, although some mothers said that they had tried to explain to these young children whey it was necessary for them to spend more time with the sick child. Parents who handled these situations most successfully were those who operated as a team with, for instance, mother giving the C.F. child physiotherapy while father was bathing the younger child. Healousies were also reduced by attempts on the part of parents to be scrupulously fair about the distribution of treats and favours although this was not always easy. Although the literature e.g. Tropauer et al, (1970) does not distinguish between older and younger siblings the demanding behaviour and the feigning of illness seemed more likely to present overt problems among these younger children e.g. several mothers described having to give physiotherapy to their healthy young children to satisfy their desires for her attention.

Among the elder siblings the situation was rather different.

Many of the mothers described their older children as being protective of their C.F. brother or sister. A willingness to defend the sick child against outside assailants was not always accompanied by personal concessions within the home when the children's interests clashed, but on the whole these older children were protective of their C.F. siblings. Being older, though, these children were more likely to have been exhorted to look after their younger siblings and so the overtly tolerant behaviour could in some cases disguise underlying resentment and it was interesting that fathers were sometimes more likely to perceive this resentment than were mothers. This may be a function of the mothers' defences against feelings of guilt about having sometimes neglected the child in some way. These defences act in such a way that the mother is more likely to describe the 'good' tolerant behaviour of the child to the C.F. child than to describe 'bad' behaviour for which she may feel herself responsible.

Certainly care is needed in the information which is given to older siblings that they should not be told anything which they could use in private assault on the C.F. child. Although mothers recorded that there was little conversation about cystic fibrosis between the children, it was not unknown for older siblings to taunt their C.F. brothers and sisters with this information. The most damaging case of this is clearly when the sibling exposes the child to the possibility that he may die. It seems that it is as disturbing for these children to be given no information as it is for them to be told of facts which they can only imperfectly appreciate, and again the nature and extent of family communication about cystic

fibrosis, including these siblings, is an important factor in their subsequent reactions, particularly if they are older than the affected child.

The sample was not large enough to allow meaningful comparisons of the social adjustment, as described by the Bristol Guide, of older and younger siblings. However, for the group as a whole there was evidence of disturbances, most severe in the over reactive mode of maladjustment, among both brothers and sisters of C.F. children. Behaviour disturbance of this nature was more evident among these children than among their matched controls or than in Stott's samples (1971). The brothers of these C.F. children seemed to present more problems than their sisters, showing inconsequential behaviour, hostility and peer-maladaptiveness in several cases. Although only a few of these children were, in Stott's terms, severely maladjusted, this is sufficient to suggest that the situation of these children has for too long been neglected, and that some attention should now be given to the situation of the healthy sibling.

A Note on Special Cases

The eight families in the sample who had two affected children seemed to represent a rather special case which was worthy of separate note.

Where the children had no healthy siblings the situation seemed to be a better one from the children's point of view in as much as the children could be treated equally within their home. Clearly if one child was much more severely affected than the

other though, imbalance in parental attention was likely to be created. From the parents' point of view, having two affected children involved a great deal more work and a much heavier time commitment in treatment which could only really be satisfactorily carried out where fathers were willing to play a regular and substantial part in treatment.

Where there were healthy siblings as well, the situation was further compounded. Four of the families in the sample fell into this category. In two of these instances the eldest child of three was healthy and was old enough to appreciate the problems facing the younger two, in an adult way. When all three children were young though, there was evidence, in a third family that an older sibling could feel very jealous if she felt that the two younger children were monopolising her parents attention. The fourth case was that of a larger family with three healthy children as well as having two C.F. children. However, one of the C.F. children was so mildly affected as to identify with the healthy children rather than with his C.F. brother so that, that situation resembled that which prevailed in families with only one affected child.

In such a small number of cases clear patterns cannot be expected to emerge. Nevertheless it is important to point out the increased demands which are made of parents who have more than one affected child, particularly if they have to attend to the needs of other healthy children as well.

The points which were made previously, referring to the development of relationships between C.F. children, apply also to the situation of the C.F. child who has a C.F. sibling.

For both relationships there are drawbacks and advantages, the only difference is that in the latter case the relationship is inevitable.

The family with more than one C.F. child has then an additional load to bear, the anxieties we have described are doubled. From this limited study it is not possible to say whether the problems are necessarily, also doubled but it seems fair to point out that these families constitute a rather special case which consequently may merit rather special attention.

The second type of family which may also be seen as constituting a special case, in a way which has not, hitherto, been sufficiently emphasised is the family where a child has already died of cystic fibrosis. Again, this study does not provide enough evidence for more than the caveat that parents responses to their living child are then at risk of being influenced more by their past experiences and their method of dealing with them than by the surviving child's existing needs. Such families too may need special help.

The Question of Stigma

From the beginning, this study set out to explore the significance of the factor of the visibility of a disability, on its influence on affected persons. Although no systematic approach was taken to this problem in the course of the study it is possible for us now, briefly, to compare relevant aspects of our findings with the observations made in the literature of the concomitants of other, visibly stigmatising disabilities.

Since not all disabilities which are later visible, are

apparent from the first, cystic fibrosis is not alone in pattern of prediagnostic events which it may present. Hewett's observation, (1970) made in relation to mothers of children with cerebral palsy, is as valid for mothers of C.F. children: "By the time that a mother is anxious enough to voice her fears that something is wrong with her baby, she needs above all to be taken seriously." Difficulties in obtaining a diagnosis are not then unique to the conditions which lack visible stigma.

The shock of the diagnosis, the lack of immediate comprehension and the need for repeated explanations are, it seems common to the diagnosis of all chronic conditions of childhood to which some permanent adaptation must be made (Spock and Lerrigo, 1965.) "Doctor-shopping" or the tendency for parents to trundle their child round several specialists in the hope of obtaining a more acceptable diagnosis, was described by Gardner (1968) in the response patterns of parents to the diagnosis of their brain-injured children. Since then, it has been said to characterise the post-diagnostic behaviour of the parents of children suffering from a variety of conditions. Doctor-shopping was rare among the parents included in this sample, and only one family was lost to the sample (after the study was complete) because of this.

The absence of doctor-shopping may be because in cystic fibrosis there is an immediate need for parents to learn as much as they can about their child's condition in order that they can take on the demanding role of supervising the child's treatment. Parents who are already committed to a programme of therapy which they can see to be effective are then much less likely to

feel the need to seek other opinions on the child's condition.

Thus, in the generalities of the early events up to and including the time of the diagnosis, cystic fibrosis may present a pattern which is comparable to that presented by other chronic conditions of childhood, but the visibility of the particular disease thereafter would seem to be an important factor in parents' long-term attitudes to the diagnosis and to the treatment which it requires.

The evidence for this last point is apparent within the findings of this study as well as by comparison with other handicaps. It was quite common for parents of children who were relatively well, graded A on Dr. McCrae's scale, to find themselves questioning the diagnosis at times, "because he/she looks so well." This response was even more common among family friends and relatives. Clearly it is fairly common for the ego-defences to deny the existence of a disability but this is a difficult position to sustain when the child is disfigured or maimed. Similarly when the C.F. child's physique is slightly abnormal or his symptoms are intrusive, there is less room for doubt or disbelief. The point to be noted, then is that the lack of visible stigma can act as an obstacle to the realistic long-term acceptance of the diagnosis by making it easier for the defences of denial and disbelief to come into operation.

In a similar way the lack of visible symptoms can make it much harder for parents to appreciate the importance of prophylactic measures. Particularly if the treatment is arduous, as physiotherapy undoubtedly is, the lack of visible reward for the parents' effort may dissuade from providing the

thorough programme of therapy which is required. The need to remind parents of the need for treatment to maintain the child's progress should periodically be observed, lest parents be lulled into a false sense of security by the child's lack of visible symptoms, and lest they thus become lax in their tratment of him.

The question of the impact of a child's disease on his family has been much stressed, particularly in terms of material hardships, social isolation and impoverished family communications. By being in possession of all his faculties the C.F. child does not place his family in the position of having to modify their home to meet his special needs, of having to buy expensive equipment or of having to consider committing the child to an institution. Thus the pressures placed on the family by the child with an internal disease are to some extent less than those imposed by most of the common visible handicaps.

The importance of visible stigmata in the social isolation imposed on families by their handicapped children was described by Kershaw (1966) who explained that with increasing visibility of the child's disability the mother's shame made her hesitate to take the child out. Some C.F. mothers described similar feelings when their children had just been diagnosed and still looked as though they were suffering from malnutrition. However the factors in the social isolation of C.F. children were fear of exposing the child to germs and the problem of social stigmata e.g. offensive odours caused by the child's stools. Whereas these were factors which might be overcome as the child became

older and stronger, and as his digestive system was better controlled, but the problems of the visible stigma were less likely to abate. From the point of view of the family's adaptation then the child whose condition is not visibly stigmatising is less likely to incur social isolation for his family. The factor of maternal anxiety in the family's social isolation is one which is common to seen and to unseen conditions.

From the point of view of communication with outsiders the factor of visibility is an important one. For the family with the child with cystic fibrosis this lack of stigmata can be both an advantage or a disadvantage. Since almost all of the children look normal when clothed and can behave in a normal way the parents, and, indeed, later the child himself can choose 'to pass', in Goffman's terminology (1968), if they so desire. i.e. they can choose to whom they reveal and do not reveal their disability. Most families make use of this to some extent. The child who lacks visible stigma need not be stared at, he can 'pass' as a normal child; he and his family are not asked thoughtless questions by the curious. However 'passing' brings its own disadvantages, the child may look normal, but his symptoms can be socially unpleasant and with no stigmatising indication of his condition they may be misinterpreted and the child may be socially sanctioned e.g. children reported having been glared at by strangers because of their rather nasty cough. Hurtful comments may be made unwittingly by those who are aware of the child's condition e.g. "who's got a little fat tummy then? Isn't it funny his tummy should be so fat when his legs are so thin?"

and Patterson's view of the public reaction to cystic fibrosis would seem to be a fair one:

> "The public renders sympathy and compassion for a crippling or chronic disease where a limp, a crutch or a brace is the symbol of an illness. The understanding that the patient and relatives receive from others observing such a handicapped child alleviates to some degree the burden by partial identification with the patient. But a child handicapped by a chronic disease which lacks such symbols receives very little empathy from the general public. In the case of cystic fibrosis the unglamarous association of foul-smelling stools, thinness, chronic cough and even disgusting purulent sputum produces a certain amount of annoyance or even loathing."

The advantages of the lack of visible stigmata in cystic fibrosis are it seems, only truly advantageous to those who are well enough to pass successfully. For those who are more severely affected, the lack of any clearly defined symbol of illness can be more of a disadvantage than an advantage.

Beyond the influence of visibility as already discussed, the important factor mediating the influence which cystic fibrosis has on parents is more likely to be the question of the prognosis. The significance of the prognostic indications will be discussed with reference to the findings from the control group, in Section VI. Nevertheless it should be recorded here that the apparent normalcy, in mental and physical terms, may be an important

factor in keeping down the numbers of parents who describe feelings of rejection of their sick child. Such feelings are commonly discussed in the literature referring to chronic conditions of childhood, e.g. by Spock and Lerrigo, (1965), by Kershaw (1966) and by Younghusband et al (1970), yet they were not found to be common among the parents interviewed in this sample. The other attitudes and emotional problems of the parents seem to be matters of common experience irrespective of whether the child's condition is apparent or not e.g. tendencies to protect the child and to be more permissive; tendencies to feel guilty for his difficulties and for the strength of the parent marital relationship to be a key issue; tendencies for parents to show health problems, for example depression. (In these respects the findings of this study may be found to be generally comparable to those reported by McMichael, 1971.)

From the point of view of the children themselves, the absence of any visible handicap is again a mixed blessing. Again, the child is not obviously barred from joinging in the activities of normal children, e.g. he can go to a normal school, he can play games and sports, but to the extent that he is not a normal child. These advantages can be disappointing in their fulfilment.

Thus, by l acking visible stigma the young C.F. child is to some extent protected from knowledge of his difference from others but when he is exposed to his own limitations, the advantages of the lack of apparent difficulties diminish. Indeed as the child becomes older such visible stigmata as are present loom larger, and are augmented by the socially stigmatising nature of

the symptoms. Thus, in the long run the problems which have to be faced by the child with cystic fibrosis are at this level, on a par with those of the visibly handicapped child. Indeed it may be argued that it is harder to bear to be so near to the goal of being physically normal and yet to fall short than it is to have to accept from the first that the goal of physical normalcy is no longer a relevant one. This discussion again ignores for the moment the question of the prognosis which undoubtedly makes the situation of the C.F. child a more distressing one than that of many of the visible but non-fatal conditions that other children may have.

The main point in relation to the question of visibility of disorder and the behaviour of the siblings is again linked to the question of communications. It may be difficult for children to understand why a brother or sister who looks just like them, gets so much more of the parents' time than is available to them. It is just as important then in these cases, as in the cases where there are visible stigma to account for, for the situation to be explained to the siblings as soon as they are old enough to understand, in a way which assures them of their own importance in the family yet conveys the special needs of the sick child.

We would submit then, that there are some aspects of the influence which illness and disability have on affected children and their families, which are mediated by the factor of the visibility of stigmatising symptoms. However, on balance, this brief overview would suggest that there are more similarities than differences between the findings of this study

and those reported in the literature to describe the concomitants of visible handicap. Visibility then does not seem to be one of the major factors in the influence which chronic illness or disability has upon affected children and their families.

SECTION V - THE CONCOMITANTS OF COELIAC DISEASE - A COMPARATIVE STUDY

The implications of the findings from the study of cystic fibrosis children and their families were evaluated with reference to a control group and this control group was selected from children suffering from coeliac disease. Although the reasons for this choice, enumerated in Section II, gave some insight into the nature of coeliac disease, we cannot expect to appreciate the situation of these families without a fuller understanding of their children's condition and its treatment. This section will provide a brief lay account of the disease before presenting the research findings.

A Lay Introduction to Coeliac Disease

As other causes of malabsorption have been recognised e.g. cystic fibrosis, so coeliac disease has, by a process of elimination, become better defined but even now its aetiology is not fully comprehended. Certainly it does not have the clearly defined pattern of inheritance of cystic firbosis and environmental factors are thought to predominate in importance in its aetiology. It had been suggested that the incidence of coeliac disease among the relatives of known cases was higher than that found in the general population but the definition of coeliac disease used at that time was unsatisfactory in the light of more recent knowledge. In 1969, Dr. McCrae reported the results of a study designed to estimate, more accurately, the incidence and

heritability of coeliac disease as it occurred in Central Scotland. He suggested that the genetic component in coeliac disease is an underlying susceptibility of multifactorial origin.

Coeliac disease, like cystic fibrosis, is caused by an internal defect. Although in its undiagnosed state coeliac disease may lead to abnormalities of physique, these are not usually gross and, at least among child patients, are normally corrected when the condition is brought under medical control. Coeliac disease then, like cystic fibrosis, lacks apparent physical stigmata.

Coeliac disease is caused by an intestinal abnormality which interferes with the absorption of nutrients from the gastro-intestinal system into the bloodstream. Although there are substantial differences in the incidence of the disease reported from different geographical areas, even within Britain it is generally agreed that coeliac disease is the most common cause of such generalised malabsorption found in temperate climates. Dr. McCrae has calculated that the incidence of coeliac disease in Central Scotland is of the order of 1 in 1,850 of the general population (McCrae, 1969).

In coeliac disease, the lining (Mucosa) of the small bowel shows an abnormal response to an unknown fraction (peptide) of wheat protein i.e. gluten. It is not known why this response occurs, although most medical textbooks seem to favour an explanation in terms of an abnormal immunological response. However, the form which this response takes, is known.

Normally the lining of the small intestine is villous, with many finger-shaped projections increasing the total of its

absorptive surface area. The effect of gluten in the diet of the person with coeliac disease is to deform this pattern in such a way that the lining appears, under the microscope, as being flat and featureless or else merely ridged. This change in morphology then interferes with the passage of nutrients from the intestines into the circulation. Although a fairly specialised knowledge of the body's chemistry is required to appreciate the exact nature of the consequences, the layman may understand the essence of the problem if he is aware of the ensuing onset of a variety of nutritional deficiencies, e.g. anaemia, because of poor absorption of iron.

Although coeliac disease may present at any age, we are here concerned only with those who were diagnosed during childhood. In that case, the symptoms of the disease usually appear after weaning, when cereals, and hence gluten, are introduced into the diet. The cumulative effect of this malabsorptive syndrome is that the child benefits less and less from the food he eats, and fails to thrive. The undiagnosed child looks not unlike the undiagnosed C.F. child, without the chest symptoms. These children then, develop pot-bellies while their limbs look wasted. Their mothers complain of their vomiting, poor appetite and greasy, offensive, loose stools. However, there is an additional symptom of coeliac disease which is a characteristic commonly mentioned; the undiagnosed children are temperamentally very trying, tending to be highly irritable and discontented.

The diagnosis of coeliac disease is confirmed by the results of a biopsy, i.e. a small piece of the small intestine is nipped

off for examination under a microscope. If the diagnosis is confirmed, a gluten-free diet is prescribed and when gluten is thus withdrawn from the diet the intestinal mucosa tends to revert to the normal villous pattern. However, this return to normal takes some time to achieve and, in the interval, dietary supplements of vitamins and iron may be given to combat the continuing tendency to nutritional deficiencies, which may otherwise result in stunted physical growth and, among girls, in infertility. Once the disease is brought under control strict adherence to this diet is usually the only treatment required.

In the past it was considered sufficient to maintain this dietary regimen until the period of growth was past. A return to a normal diet in adulthood was clearly more convenient for the patient and was thought unlikely to be prejudicial to his health by that stage. However, it has since been observed that the incidence of malignancies developing in later life in the gastro-intestinal tract, is higher among such untreated coeliac patients than among the general population. In order to avert increasing the risk of malignancies then, the gluten-free diet has come to be regarded as a life-long treatment for coeliac disease.

The Treatment of Coeliac Disease

It is no mean feat to eliminate gluten entirely from the British diet. All foods made with normal flour become taboo; special gluten-free flour is available on prescription and it must always be used in its stead. This flour has rather different characteristics from normal flour and although mothers are encouraged

simply to use it as a substitute, the results can be very disappointing, in some cases.

Some commercial bakers will undertake to make the special bread which the children require but it is not always very palatable, it can be rather expensive and its keeping properties are poor. A few varieties of plain biscuits are also available on prescription but beyond these things the onus is really on mothers to master the art of cooking with this flour.

The problem does not end with the obvious flour-based foods. Wheat starch and gluten-based compounds are common additives to many proprietary brands of tinned and frozen foods, and they occur in common foodstuffs, such as sausages, and in some brands of confectionery. It thus requires a high degree of vigilance to achieve and maintain a gluten-free diet.

After the child's diagnosis mothers are usually provided with a booklet advising them of the foodstuffs which are and are not included in this diet. They are then asked to bring the child to the Outpatient Clinic, the same one as is attended by the C.F. children, at regular, usually monthly, intervals. At these visits the child's growth and development are assessed and the adequacy of the control of his diet is discussed. The clinic in Edinburgh is fortunate in having the voluntary services of a lady, herself the mother of two thriving coeliac children, who is able to give sound advice to mothers who have difficulties with the prescribed diet. When satisfactory control has been established, i.e. the children show an improvement in their weight gain, growth pattern and physique, a more normal bowel

habit and an improvement in their general wellbeing, then they may settle to a routine of only a few clinic visits per year. This pattern is usually then maintained throughout childhood, at least in the Royal Hospital for Sick Children in Edinburgh, in order to check that progress is being maintained. Relapses which occur are nearly always the consequence of the dietary rules being broken.

Clearly then, this diet can be socially most inconvenient for these children and their parents. There is a Coeliac Society which endeavours to help alleviate some of the problems and its role will be discussed in Part D.

For the moment, we are concerned to identify the extent of the influence which this disease has on these families and to assess whether, as another example of a chornic disease of childhood, it presents a situation for families which is significantly different from that posed by cystic fibrosis and if so, why this should be.

As a control study, this investigation was primarily designed to provide information for comparison with the data collected from C.F. families. However, the study of the social and psychological concomitants of coeliac disease was also interesting in its own right, since a survey of the literature failed to reveal any previous research in this field. The one study reported in the literature, by Prugh (1951), showed many of the methodological shortcomings already noted in earlier studies of cystic fibrosis, e.g. Prugh's sample size was very small and very sweeping generalisations were made on the basis of very little evidence. However, the most damning factor, which

effectively renders it irrelevant to this study, is that it was written at a time when coeliac disease was not well understood. The course of the disease and its treatment, as described by Prugh bear very little resemblance to the situation described here. Prugh's thesis, on the basis of a study of only 14 cases, was that there was, both among coeliac children and their mothers, a "consistent though non-specific personality type and conflictual background", which was said to be influential in the onset and course of the disease in the children.

Prugh's work then, was of little help to our investigations and the study of coeliac children and their families followed the same pattern as the study of C.F. children, with modifications introduced only to explore the treatment of coeliac disease. The findings of this control study are therefore presented and compared with the findings from the C.F. group to which it was matched. The order of presentation of these results is, as before:

A. Family Settings.

B. Diagnosis and Treatment.

C. Family Functioning.

D. Chronic Illness in Childhood - The Effect on Parents.

E. Chronic Illness in Childhood - The Influence on the Development of Affected Children.

F. Healthy Siblings of Chronically Ill Children.

(The results quoted in these chapters are derived from interview schedules which varied little from those used in the C.F. Study. The schedule for parents and that for children were

modified in some respects to take account of the different treatment procedure involved and these modified schedules appear in Appendix V. However, the numbers of the questions have been kept the same as in the original version so that the references given in Section III still hold.

The individual interviews for mothers and fathers were not modified and the reader is referred once more to Appendices II and III for the relevant schedules. Here, the only change to be made is the substitution of the words 'coeliac disease' for those of 'cystic fibrosis'.

A. Family Settings.

In gathering background information about the children with coeliac disease, for comparison with the C.F. children, we were deprived of one of the previously most important variables, the index of the child's clinical condition. Although in coeliac disease, varying degrees of tolerance of gluten in the diet do seem to be reported by the children's mothers, clinically, coeliac disease is regarded as an all-or-none condition which is not amenable to scaling for severity as cystic fibrosis is. Consideration of the child's physical condition is therefore omitted altogether from this section.

Comparable background information was available, however, about the other aspects of the families' circumstances, in terms which have already proved useful to our understanding of the concomitants of cystic fibrosis. First, however, it is necessary to clarify the nature of the sample of coeliac children and their families which is to be described here.

It will be recalled from Section II that this control sample was derived to provide 54 coeliac children, matched in several important factors to 54 of the C.F. children. As far as possible then, the results in this Section will be reported in terms of the families of the 54 children, in each of the two samples. However, since some families have two C.F. children and since complete records are not available for all parents, the two samples do not represent data from equal numbers of parents. This need not be a serious disadvantage to research of this kind for, by expressing the data in percentages, we may still obtain an impression of the comparability

of the findings, simply by observation. The exact numbers of individuals or families from whom the data were derived will be indicated, and, as far as possible the coeliac results will be presented with the corresponding findings for the matched C.F. sample alongside.

Parental variables of age, level of educational attainment, occupation and marital status are quoted in this way. These data, along with information about family composition and housing, are recorded in this chapter to enable the findings relating to the coeliac families to be evaluated in their proper perspective, for they may represent important sources of uncontrolled variance in the results reported.

The parents in the two samples were very similar in age Table V.A.(i). Biographical information having been gathered from mothers, we are enabled in this chapter to describe more fathers than the number who contributed personally to our study.

Table V.A.(i). The Ages of Parents in the Two Samples

		Sample	
		Coeliac	C.F.
Mothers :	N	54	48
	Age range	24 - 51 yrs.	21 - 53 yrs.
	Mean age	33.6 yrs.	33.0 yrs.
	S.D.	6.0	7.5
Fathers:	N	51	48
	Age range	24 - 54 yrs.	21 - 68 yrs.
	Mean age	36.5 yrs.	36.5 yrs.
	S.D.	7.3	8.5

Of the three missing fathers indicated in the coeliac sample one had died, one was divorced and his former wife could not remember his age and the third represents an unmarried mother.

The number of years of formal education undergone by the parents was similar in the two groups. On average coeliac mothers had left school at the age of 15.3 years, and C.F. mothers at 15.2 years. Coeliac fathers' education had ended, on average, at the age of 15.4 years, while that of C.F. fathers had ended slightly earlier, at 15.1 years. These differences are not significant. Again, normal distributions of scores on Cattell's factor B, for mental capacity, were observed among parents in both groups.

The patterns of mothers' employment at the time of the enquiry suggested a tendency for mothers of C.F. children to be less likely to go out to work (Table VA.(ii)).

Table V.A.(ii). The Employment Status of Mothers of Coeliac and C.F. Children

	Coeliac	C.F.
In full-time employment outwith home	6%	4%
In part-time employment outwith home	35%	22%
In part-time employment at home	-	6%
No gainful employment	59%	68%
	N = 54 mothers	N = 47 mothers

The proportion of mothers taking night-time employment was mentioned previously when 8 of the 14 C.F. Mothers who held part-time jobs were observed to go out to work at night. Most of them held jobs which allowed them to return home later the same night e.g. office-cleaner, barmaid, although one of them was a nurse who worked on night-duty throughout the night for a few nights each week. Only two of 19 part-time workers among the coeliac mothers went out to work at night and both of them were night-nurses. Cystic fibrosis then seems to present, not surprisingly, more of an obstacle to mothers wishing to take up some form of outside employment than does coeliac disease.

It is interesting then to consider the pattern of employment of fathers. Table V.A.(iii).

Table V.A.(iii) Fathers' Presence in the Home

	Coeliac	C.F.
Unemployed, retired	4%	12%
Normal working day	62%	49%
Away from home several nights/week	6%	16%
Away from home regularly	13%	9%
Away from home for long periods	2%	4%
No father in the home	13%	10%
	N = 54 families	N = 48 families

Clearly there are some slight differences in the pattern of employment of the fathers of children in the samples. Not too much significance should be attached to these differences at present,

although they should be borne in mind in the course of the interpretation of related findings. The relatively higher unemployment figure among the fathers of C.F. children may mirror the national trends which showed higher unemployment figures in the country as a whole at the time of that part of the study and which had begun to improve by the time of the coeliac study.

A consequence of chronic illness among children which was frequently described in the literature was that of breakdown in marital relationships. Separation and divorce rates are often reported to be higher among the families under such stress. It is interesting then to compare the structure of the families included in the two samples of this study. The marital status of the parents of these children is indicated by Table V.A.(iv). The figures quoted for separations are slightly inaccurate as two further couples, parents of C.F. children had separated shortly after making their contribution to the study whereas two sets of parents of coeliac children had become reconciled.

Table V.A.(iv). The Marital Status of Parents of Children in this Study.

	Coeliac	C.F.
Normal marital status	81%	81%
Parents separated/divorced	9%	13%
Parents did not marry	4%	4%
Other,(e.g. mother widowed)	6%	2%
	N = 54 Children	N = 54 Children

This information about the structure of the relationships within the children's families is less important in this Section than in Section III however, since the question of genetic counselling in relation to coeliac disease does not arise.

Nevertheless it was still important for the study to assess the relative impact of these two chronic diseases on marital stability so that note was still taken of the duration of parents' marriage as a possible factor in its stability. Here again however, the differences between the two groups were minimal. The parents of the coeliac children had been married for an average of 11.4 years (S.D. = 5.5) while the parents of the corresponding sample of C.F. children had been married for an average of 11.0 years (S.D. = 6.8).

The mean family size of these families has already been recorded in Section II as one of the bases for the selection of the control group. However it is interesting to point out here that the reproductive histories of the families in the two groups show significant differences. Whereas one coeliac family had lost a child, in an accident, and two mothers had suffered, between them, four miscarriages, 10 of these 48 C.F. families had lost a total of 13 children and 16 of the mothers had had, in all, 24 miscarriages. Had all their children survived then, these C.F. families would have been, on average rather larger than those of the control group.

As in Section III, so here it is important to view the families against that home background and to compare the two groups in this respect. Since the child with coeliac disease was not felt to impose any special housing requirements, only

two of the factors discussed previously are included in this Section i.e. overcrowding and the adequacy of the state of repair and facilities of the families' homes.

Using Doulgas and Blomfield's index of overcrowding (1958), that is, designating as overcrowded any living conditions which represent a density greater than $1\frac{1}{2}$ persons per room, 19% of these coeliac family homes were overcrowded. Not only was this overcrowding slightly more serious than among the 16% of C.F. families, who were also said to be living in overcrowded conditions, but it was more permanent. Unlike the C.F. cases previously reported, none of these coeliac families had any moves afoot to reduce the pressures on their accommodation. This situation may have been exacerbated by the tendency to larger families among the coeliac group.

In the control group too, the proportion of home owners was higher than among the C.F. group, Table V.A.(v), and this might also be a barrier to spaciousness of their homes.

Table V.A.(v) Housing Status of Families in the Two Groups

	Coeliac	C.F.
Owner-occupiers	43%	25%
Tenants	50%	69%
Other	7%	6%
	N = 54 families	N = 48 families

The adequacy of the state of repair of these homes was assessed by observation as before, and it tended to be slightly

lower than the standard achieved by the homes of the C.F. families. The mean index value obtained by the control group houses was only 8,9, S.D. = 2.6, as compared with the mean value of 9.4 and standard deviation of 2.0 obtained by the C.F. families. These scores were derived from cumulated totals of scores assigned to four aspects of the family's accommodation as before (Table III.A.(v)). If the score assigned was less than 8 out of the possible score of 12 i.e. no aspect scored better than 'moderate', then it was likely that the mother of that family would also consider that her home did not adequately meet her family's needs (Coeliac, $X^2 = 8.80$, d.f. = 1, $P < 0.01$; C.F. $X^2 = 14.55$, d.f. = 1, $P < 0.001$).

The families in the two groups were alike in the duration of time they had spent domiciled in neighbourhood in which they lived at the time of the investigation. All but seven families in each group had lived in their present homes for more than a year before taking part in this study. Only 5 coeliac mothers and 3 C.F. mothers actively disliked the vicinity in which they lived, and although some mothers had mixed feelings about their neighbourhood, 68% of mothers in each group were quite happy with the situation of their homes.

On the whole then, it may fairly be said that the families included in the two samples were very similar in the relevant aspects of their background, even though no active steps had been taken to control for these factors. It is hoped that this similarity between the groups may allow a useful comparison of their results to be made in such a way that the uniqueness or generality of the findings reported from the C.F. families may be better evaluated.

B. Diagnosis and Treatment

Issues concerning the medical management of cases of cystic fibrosis have already proved essential to our understanding of the implications of that disease for affected children and their families. Similarly, the value of the control group is clarifying these findings still further, also depends on the provision of an accurate picture of the medical background against which the data may be better interpreted.

Although coeliac disease may appear among the differential diagnoses in a case of cystic fibrosis, and perhaps, vice-versa, the clinical aspects of the comparability of these two conditions should not be stressed, since differences between them, in these terms, considerably outweigh their similarities. However, this thesis is conferned with psychological concomitants and not with clinical features per se, and since both conditions represent instances of chronic, incurable, astigmatic diseases of childhood, the literature leads us to expect to find a degree of commonality of experience, even in these essentially medical matters of diagnosis and treatment. On the other hand, to the extent that the factor of their differing prognoses is influential we should also be prepared to find some significant differences in the experience of the two groups.

As far as possible the data from the two groups will be presented in parallel, for easy comparison and it will be considered in the same order as in Section III.

i.e. 1) Diagnosis

2) Parental Comprehension

3) Medical Supervision

4) Treatment

5) Agents of Help

6) Hospitalisation

1. The Diagnosis

The children in the two samples had, on average, had their diseases diagnosed at approximately the same ages. The mean age of diagnosis among the 54 coeliac children was 23 months, (1:11 years) with a standard deviation of 27.8. The mean age of diagnosis of cystic fibrosis in the matched group of 54 children was 24 months (2:0 years) with a standard deviation of 30.5.

81% of the coeliacs and 78% of the C.F. children had been diagnosed in the Royal Hospital for Sick Children in Edinburgh. 52% of all the coeliacs and 48% of all the C.F. children in the study had been diagnosed by Dr. McCrae.

Mothers of coeliac children were asked to provide a brief history of their prediagnostic and diagnostic experiences. The events recounted allowed a coded assessment to be made, as before, of the degree of difficulty which these mothers had encountered in trying to obtain a diagnosis for their children. Although this coding scheme operates on the same principles as that used to describe the difficulties experienced in trying to obtain a diagnosis of cystic fibrosis, the events which it summarises are

slightly different and specific illustrations are quoted in explanation:

Great difficulty. e.g. From the age of 0:3 C. was sick after every feed. This vomiting and diarrhoea persisted for several days so C's mother consulted her G.P. He diagnosed gripe and prescribed a bottle of medicine. It was ineffective and the mother returned repeatedly to the doctor's surgery. She felt he thought that she was fussing about nothing but he did continue to prescribe a succession of medicines, each as ineffective as the previous one. By the age of 0:9 the mother was alarmed because the child was 'wasting away' and finally she insisted on the child being referred to hospital. The child was admitted and discharged again after a week of tests. The mother was told there was nothing wrong. After six more weeks the child was still not thriving and the mother, by then very anxious, begged for a second referral to the specialists. On this second occasion the child was admitted to hospital for four weeks, after which time coeliac disease was diagnosed. C was 1:0 year old and very ill.

Some difficulty: At about 0:4 yr. S. began to have solid food. Shortly afterwards he had a bout of diarrhoea and vomiting which the family doctor diagnosed as gastroenteritis. He recommended that the child be starved and given only water for a day or two. Liquid feeding was then to be resumed and solids reintroduced only when S. showed signs of recovery. Shortly after returning to solid foods S. had diarrhoea and vomiting and the doctor offered only the same recommendation. By modifying the child's diet S's mother was able to find some foods which did not upset

him, so that S continued to gain weight, but, by the age of 0:9 yr. he was still having recurrent gastrointestinal problems. His mother, not satisfied that the child had gastroenteritis, sought a second opinion from the family doctor of her relatives. Through him the child was admitted to the local hospital and, shortly afterwards was transferred to R.H.S.C. S. was diagnosed as having coeliac disease at age 1:0year.

Little or no difficulty: B. was not thriving. On two consecutive visits to the Local Authority Baby Clinic he had failed to show any weight gain. His mother described his bouts of diarrhoea and vomiting and was immediately referred to her G.P. He referred the child to hospital and B was diagnosed as having coeliac disease at 0:8year.

The experience of at least some difficulty in obtaining a diagnosis for their children, was not then peculiar to the mothers of cystic fibrosis children, although, admittedly, they were more likely to describe greater difficulties than the mothers of coeliacs. Table V.B.(i).

Table V.B.(i). The Incidence of Difficulty Experienced in Obtaining the Diagnoses.

	Coeliac	C.F.
Great difficulty	24%	41%
Some difficulty	46%	22%
Little or no difficulty	30%	37%
	N = 54 diagnoses	N = 54 diagnoses

The mean time interval over which these events were recalled was 5:5 years in case of the control group and 5:2 years since the diagnosis of the C.F. children.

In view of the similarity of the presenting symptoms of the two conditions in many cases it was perhaps rather surprising that the difficulties described by these coeliac mothers did not reflect the mothers age at that time, as they id in the cases of cystic fibrosis previously described. Table VB.(ii). However, two other points suggested by the cystic fibrosis mothers, but not found to be significant in their influence in that sample, were found to be associated with difficulties in obtaining a diagnosis in cases of coeliac disease. Table V.B.(ii). Among the coeliac families then, greater difficulties were more commonly described when the child was the first-born and when the family was of lower social class status. In neither the coeliac, nor in the C.F. sample, was there any significant evidence that the experience of such difficulties had lessened in recent years.

The pattern of blame attributed by the mothers for the delays in the diagnosis of their children, was similar in both groups, with family doctors coming in for most criticism. Table V.B.(iii).

Table V.B.(ii). Factors Relating to Mothers' Difficulties in Obtaining Diagnoses.

	Coeliac	C.F.
Age of Mother at Diagnosis	$X^2 = 0.81$ d.f. = 2; $P \simeq 0.70$	$X^2 = 9.12$ d.f. = 2; $P < 0.02$
Birth Rank of Child in Family	$X^2 = 3.75$ d.f. = 2; $P \simeq 0.20$	$X^2 = 1.55$ d.f. = 2; $P \simeq 0.50$
Socio-economic Status of Family	$X^2 = 4.68$ d.f. = 2; $P < 0.10$	$X^2 = 5.99$ d.f. = 2; $P < 0.05$
Time lapse since Diagnosis (< 4 years vs. ≥ 4 years)	$X^2 = 1.72$ d.f. = 2; $P \simeq 0.50$	$X^2 = 2.82$ d.f. = 2; $P \simeq 0.30$
	N = 54 diagnoses	N = 54 diagnoses

Table V.B.(iii). Incidence and Direction of Blame for Delays in Diagnoses.

	Coeliac	C.F.
Blames self	11%	7%
Blames family doctor	35%	28%
Blames hospital doctor	17%	22%
	N = 54 diagnoses	N = 54 diagnoses

Since these categories are not mutually exclusive, and do not include those mothers who apportioned no blame, the column values do not sum to 100%.

Lasting changes in mothers' attitudes to the medical profession, as a direct result of these experiences, were more commonly described among C.F. mothers, where 17% of the mothers described a loss of confidence in doctors and where 15% expressed greater faith in them after their experiences at the time of their child's diagnosis. Only 11% of the coeliac mothers described more negative attitudes and only 4% had felt an attitude change in a positive direction, since the diagnosis of their children.

Previously it was noted that the circumstances of the communication of the diagnoses of cystic fibrosis considered in this Study, had not been ideal. It is important to assess the extent to which these circumstances are unusual, and for the remainder of this chapter, the findings relating to the management of the diagnosis of coeliac disease and to the parents' reactions to this diagnosis will be compared with the C.F. results, to clarify this point.

Once more, it was of interest to learn which member of the family had been the first recipient of the child's diagnosis. Table V.B.(iv).

Table V.B.(iv). To Whom was the Diagnosis First Given.

	Coeliac	C.F.
To mother only	81%	44%
To father only	15%	13%
To both parents	4%	37%
Other	-	6%
	N = 54 diagnoses	N = 54 diagnoses

It seems then that the proportion of cases of cystic fibrosis, in which both parents were given the diagnosis was, in spite of our earlier criticisms, already a considerable advance on the proportion of diagnoses of coeliac diagnoses given to both parents. Of course it may reasonably be argued that this situation is more tolerable for the communication of a diagnosis of coeliac disease, where the implications are less grave and the dietary treatment prescribed falls much more heavily into what is traditionally considered to be the mothers' province.

It was interesting then that coeliac mothers seemed more likely to take advantage of available opportunities to further their understanding of the child's disease, on subsequent occasions. Table V.B.(v).

Table V.B.(v). Subsequent Discussions re: Diagnosis.

	Coeliac	C.F.
Diagnosing physician apparently unavailable	24%	28%
Physician available; Parents did not seek to re-open discussion	7%	22%
Physician available; Parents sought further discussion	69%	50%
	N = 54 diagnoses	N = 54 diagnoses

In the light of advancing medical knowledge, it was to be expected that the original information given to mothers of coeliac

children at the time of the diagnosis would, in some cases, have had to be updated. The task of the study was then to endeavour to assess what information parents had about the condition at the time of the study. Thus, parents were asked whether it had been suggested to them that coeliac disease might be an inherited condition, at least in part; whether they had been told that the child would always require a gluten-free diet and finally, whether they had been told of any risks which might be introduced by continued waiving of these dietary rules. On this basis, the themes of inheritance, chronicity and prognosis, already assessed in relation to cystic fibrosis, could be identified. The corresponding findings are then presented in Table V.B.(vi).

Table V.B.(vi). Parents' Information Re: Their Children's Diseases.

	Mothers		Fathers	
	Coeliac	C.F.	Coeliac	C.F.
Inheritance	59%	89%	39%	78%
Chronicity	59%	100%	68%	91%
Prognosis	7%	57%	7%	51%
	N=54	N=47	N=44	N=45

From parents' responses to the open-ended question about what they had been told by the doctor at the time of the diagnosis, it seemed that communications of these two diagnoses differed tremendously in approach. Whereas the coeliac parents described

an emphasis on the need for dietary restrictions i.e. on treatment, the C.F. parents were clearly given relatively more explanation about the nature and cause of their child's disease. These responses were then mirrored in the findings of the Table above (V.B.(vi)) which suggest that the C.F. parents have considerably more information about their child's condition than do coeliac parents.

Since we are concerned, not only with the information-content of diagnosis communication, but also with parents' attitudes to the manner in which the communication was made, it is of interest to compare parents' appreciation of trouble taken to explain the diagnosis to them in these two groups Table V.B.(vii).

Although the proportion of parents of coeliac children who felt that trouble had been taken, was smaller than that of C.F. children, the degree of overall satisfaction was very similar in the two groups. 46% of coeliac mothers and 43% of C.F. mothers described complete satisfaction with the management of their child's diagnosis.

Table V.B.(vii). Trouble Taken in Explanation of Diagnosis to Parents.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
By No-one	54%	45%	69%	59%
By Hospital Doctor	31%	49%	19%	39%
By Family Doctor	11%	8%	12%	6%
By Others (e.g.Pharmacist, Dietitian)	11%	4%	2%	2%

Refers to N = 54 Diagnoses of C.F. and of Coeliac Disease

Since the proportion of mothers expressing only positive attitudes about the management of their children's diagnoses represented less than half of the sample, it did appear that there was some room for improvement. It was necessary, then to probe a little further into parents' attitudes to their experiences at this time.

C.F. parents, it will be recalled, had been critical of explanations of that disease which were given either in hopelessly gloomy or in unrealistically euphemistic terms. For coeliac parents the difficulty was simpler; they were critical only of delays in the explanation of the child's diagnosis and of communications which described only the treatment, without explaining why it was necessary.

All the parents in the study were asked whether they felt doctors should tell parents what they suspect to be the matter with their child or whether they should maintain silence until their suspicions have been confirmed. The responses of parents from the two groups are compared in the next Table (Table V.B.(viii)). Clearly what the doctor tells his patients (or the patient's parents) and when he does so, must remain a matter for his professional judgment. However, it is interesting to consider the implications of his decision for those who are the recipients of his communications.

Table V.B.(viii). Parents' Views of the Ideal Timing of Diagnostic Communications.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
When doctor suspects	57%	51%	61%	31%
When doctor is sute	31%	47%	32%	56%
Don't know	12%	2%	7%	13%
	N=54	N=47	N=44	N=45

Once the prediagnostic stage is over and the diagnosis has been made, the common ground between the two groups of families diminishes. Although 25% of the mothers of the coeliac children described their shocked reactions to the knowledge that their child had a chronic and incurable disease and although 41% of mothers in that group admitted a dominant fear that their child was about to die, none of them really had to face the flood of intense affects experienced by the parents of C.F. children. By the time that most C.F. parents were undergoing a stormy passage through a period of anticipatory mourning, the parents of coeliac children were preoccupied with lesser anxieties about how they were going to cope with the child's diet. Such practical worries were described by 35% of these mothers. These findings are in marked contrast to the 78% of C.F. mothers whose abiding fear at that time was that the child's death was imminent. Only 16% of these C.F mothers said that they had primarily been anxious about their own ability to cope.

The active search for information is said to be a natural product of parents' attempts to come to terms with their situation in cases such as these. It is perhaps surprising then that parents of C.F. children were more likely to experience the impetus to seek out further information than were their coeliac counterparts, even though a sizeable minority of the former were afraid of what they might thus discover. Half of the coeliac parents and two-thirds of the C.F. parents in the study had engaged in a n active search for further information. A continuing desire for further information, even at the time of the enquiry, was expressed by 37% of the coeliac parents and by 57% of the parents of C.F. children.

The proposition that the diagnosing physician might, in future, provide parents with some written information to take home with them after the diagnosis, was accepted with enthusiasm by 80% of parents in each group.

It was of interest to learn how parents assessed the effectiveness of a variety of agents of help available to aid them in their efforts to comprehend their child's disease. As before, the sources are ranked in order of decreasing helpfulness, in Table V.B.(ix), with the mean score attributed to them quoted in parentheses. (For a fuller explanation see Section II.B.1).

Table V.B.(ix). Aids to Parental Understanding of Diagnoses

MOTHERS:	Coeliac		C.F.
Clinic doctors	(2.0)	Clinic doctors	(2.4)
Dietitian	(1.3)	Family doctor	(1.4)
Books, Family Doctor	(1.0)	Physiotherapist	(0.9)
		Books	(0.8)
Coeliac Society	(0.9)	C.F. Parents' Group, Other Parents	(0.7)

FATHERS:	Coeliac		C.F.
Books	(1.4)	Clinic doctors	(2.2)
Clinic doctors	(1.0)	Books	(1.2)
Family doctor, Coeliac Society	(0.8)	Family Doctor	(0.8)
		C.F. Parents' Group, Other Parents	(0.6)

In view of the findings reported in this chapter it is interesting then to compare the distribution of the indices of parental satisfaction with the management of their child's diagnosis obtained by each group. The index was derived as indicated in Table III.B.(viii). From the reports of coeliac parents values of the index ranged from 0-14 and showed a mean value of 6.4 and a standard deviation of 3.5. On the same terms, the index values describing the 54 C.F. diagnoses ranged from 1-17 and showed a mean value of 9.2 with a standard deviation of 3.6. The difference between these mean values was found to be significant at the 0·1% level ($t = 6{\cdot}32$)

In neither group did this index of satisfaction distinguish more recent from earlier diagnoses (Coeliac, $X^2 = 0.04$, d.f. = 1, $P \triangleq 0.80$; C.F., $X^2 = 0.23$, d.f. = 1, $P \triangleq 0.70$) nor diagnoses made in Edinburgh from those made elsewhere (Coeliac, $X^2 = 1.18$, d.f. = 1, $P \triangleq 0.30$; C.F., $X^2 = 1.31$, d.f. = 1, $P \triangleq 0.30$). The adequacy of the management of the diagnosis, as indexed here, is a factor which reappears throughout this Section. For the present, the remainder of this chapter is concerned with aspects of the long-term implications of these diseases for affected children and their families.

2. Parental Comprehension

The whole issue of the amount of information which people have about the medical conditions which affect their lives is an important one in the terms of this study. As in the study of cystic fibrosis families, so here too, coeliac parents had been asked several questions to tap the extent of their comprehension of their child's disease. The level of their understanding was graded 'good', 'moderate' or 'little or none' as before, on the basis of their responses. It was rather harder to describe objective criteria for this assessment than in the C.F. study. In both cases the investigator, a lay person, had acquired a degree of understanding of the disease in question which, in its essential elements was within the grasp of most parents. Parents who, with their experience of their sick child, showed a degree of understanding compatible with that of the investigator were said to exhibit good understanding. Where a fundamental gap was discerned in an otherwise good appreciation of the disease, e.g. understanding the nature of coeliac disease and its treatment but not accepting that the diet would always be required as treatment, understanding was scored as being 'moderate', 'Poor'understanding was readily identifiable, characterised by complete ignorance or several basic misconceptions. Using such a classification scheme it was possible to compare the degree of comprehension shown by parents in the two groups, of their children's diseases. Table V.B.(x).

Table V.B.(x). Parental Comprehension of Disease

Rating	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Good	52%	38%	48%	31%
Moderate	35%	43%	27%	22%
Little or none	13%	19%	25%	47%
	N = 54	N = 47	N = 44	N = 45

On average then parents of coeliac children were more likely to acknowledge awareness of salient points of information about their child's disease than were the parents of the C.F. children. In view of the time which they spent caring for and treating their children it was not surprising that mothers should show a higher degree of understanding of their diseases than fathers. Some of the factors which were found to be related to the extent of parents' understanding are tabulated in Table V.B.'xi). Assuming understanding to be a continuous variable a Pearson product-moment correlation co-efficient was calculated to describe its relationships with these factors. Significance values are given below each value of r, in parentheses.

Table V.B.(xi) Some Factors Associated with the Level of Parents' Understanding of Child's Disease (Pearson's r)

		MOTHERS		FATHERS	
		Coeliac	C.F.	Coeliac	C.F.
School leaving Age	r=	0.34	0.45	0.42	0.40
	p=	(0.006)	(0.001)	(0.001)	(0.002)
Intelligence (Cattell's B)	r=	0.26	0.19	0.28	0.16
	p=	(0.04)	(0.09)	(0.03)	(0.14)
Index of Diagnosis Management	r=	0.22	0.11	0.42	0.34
	p=	(0.05)	(0.22)	(0.001)	(0.008)
		N=54	N=47	N=44	N=46

Since the parents in the two groups were not significantly different in terms of their intelligence or years of formal education, these were not thought to be the factors implicated in their differential levels of comprehension (Table V.B.(x)).

The mean index values describing the adequacy of the management of diagnoses in the two groups would have suggested that the C.F. parents were, rather, better equipped to comprehend their situation than were the parents in the control group.

Two main reasons for the relatively poor level of comprehension shown by the C.F. parents, suggest themselves. In the first place there is a good deal more information to be assimilated by these parents and in addition it is much more

threatening in its content. The combined factors of complexity and the operation of the defences would seem to explain these parents' difficulties and to point out once again that there is still a need for further improvement in the management of the communication of the diagnosis of cystic fibrosis to parents.

In diseases such as cystic fibrosis and coeliac disease, much of the responsibility for the child's clinical progress rests with his parents. Their successful maintenance of the prescribed programme of home therapy depends, in turn, on their appreciation of the nature of the disease and the rationale of the treatment procedure.

In cystic fibrosis, it will be recalled there were additional genetic reasons for ensuring that parents understood fully the implications of their child's disease. There was no such justification for probing the matters of family planning or genetic counselling with the parents of coeliac children so no comparable data available to correspond with those of Section III.B.2.

Mothers of coeliac children were asked only whether they felt that having had one child with coeliac disease had altered their plans for further children. Only 6% of these mothers were very much against having another child lest it too should have coeliac disease. 18% said that had they realised there was a risk to their subsequent children they might have been deterred but their responses were entirely hypothetical. The remaining 76% of mothers as anticipated said that they would not be deterred, although it was interesting that they almost all qualified their responses in terms of their own ability to cope,

rather than in terms of risks or of the problem for the child. The concept of 'burden' again then seemed to be an important one in the reproductive attitudes of these mothers.

In terms of the provision of medical supervision for cases of coeliac disease we return once more tha n area of common ground between the two groups and mothers attitudes to the system of care provided are compared in the next part of this Section.

3. Medical Supervision

Even after the disease had been diagnosed and the appropriate treatment routine had been instituted, the development of most of the children in this study remained under fairly close medical supervision.

It was interesting to consider first, the extent to which prediagnostic experiences and post-diagnostic relationships with specialists, had usurped the role of the family doctor in the care of the child.

The mothers of each of the children in the study were asked firstly to whom they would refer for help or advice about the child's conditions. 81% of the mothers of coeliac children were quite content to turn to their general practitioner in the normal way but it was evident that the attitudes of mothers of C.F. children were rather different, since only 56% of them said they would refer to their G.P. The remainder of the mothers in both groups referred directly to the hospital physicians, excepting one mother in the C.F. group who tended to consult her local pharmacist.

The frequency with which such consultations may arise can be seen in Table V.B.(xii) where the frequency of mothers' visits to family doctors and clinics are compared. Once again the frequency of mothers' contact with their G.P.'s was a reflection of the age of the child in question, younger children tending to present more frequent minor problems. Since the problems of achieving satisfactory control over the condition of a coeliac child are rather less than those presented by cystic fibrosis it is not really surprising that, in spite of their greater readiness to consult their G.P.'s coeliac families do not in fact, need to do so any more frequently than do the C.F. families in this sample.

Table V.B.(xii) Frequency of Contact with Doctors.

	G.P.		CLINIC	
	Coeliac	C.F.	Coeliac	C.F.
1/month - 1/2 months	26%	24%	13%	81%
1/3 months - 1/6 months	69%	63%	81%	11%
1/6 months	5%	13%	6%	8%
	(N = 54 children in each case)			

Mothers' attitudes to their family doctors were indexed as before (Table III.B.(xvii)) and the distribution of values obtained are given in Table V.B.(xiii). The t-test of the difference between their means gave a value of t = 1.72, which was significant only at the 10% level.

Table V.B.(xiii) Distribution of Indices of Attitudes to G.P.'s.

	Coeliac	C.F.
Index Value		
0 - 1 (negative attitudes)	37%	24%
2 - 4	41%	41%
5 - 6 (positive attitude)	22%	35%
Mean index Value:	2.7	3.2
	N = 54	N = 54

The social class difference in attitudes to family doctors observed among C.F. families was less evident in the control group (Coeliac, $X^2 = 2.91$, d.f. = 2, $P \simeq 0.20$; C.F., $X^2 = 10.34$, d.f. = 2, $P < 0.01$) but those who held favourable towards their G.P. were more likely to consult him first if they had a problem with the child (Coeliac, $X^2 = 5.08$, d.f. = 2, $P < 0.10$; C.F., $X^2 = 18.58$, d.f. = 2, $P < 0.001$) and to maintain more regular contact with him (Coeliac, $X^2 = 3.53$, d.f. = 2, $P \simeq 0.20$; C.F., $X^2 = 14.42$, d.f. = 2, $P < 0.001$). The stronger relationships shown here between the attitudes and behaviour of the C.F. group are rather interesting. It will be recalled that although C.F. mothers seemed to have greater difficulties in obtaining their child's diagnosis, fewer of them seemed to blame the family doctor for these difficulties and on the whole they expressed more positive attitudes towards their family doctors than did the coeliac mothers. In spite of their more frequent

medical supervision from the out-patient clinic, the frequency of contact between the C.F. mothers and the family doctor was almost the same as that of the coeliac children. Although it could of course be argued that the medical needs of the C.F. child are greater these findings taken together suggest that in spite of his lack of specialist knowledge the family doctor has an important role to play for some of these mothers, providing a more personal source of reassurance for them than they perhaps find in the hospital out-patient clinic.

In view of the reduced frequency of their contact with doctors there was less opportunity for coeliac mothers to experience problems of communication between the clinic and the family doctor. Where problems did arise, they usually concerned the G.P.'s doubts about prescribing foodstuffs and they were relatively easily remedied. Accordingly then, 69% of the mothers in the coeliac group described no such problems. This was in contrast to only 46% of the mothers of C.F. children who had not experienced any such difficulties. The remainder of these mothers, it will be recalled had experienced long delays between their clinic visit and the arrival of the advisory letter from the hospital to the general practitioner, and on occasion, the problem had been compounded by confusions or disagreements between the two about what was to be prescribed. Clearly this system holds more hazards for C.F. families than for those with coeliac children.

In terms of the provision of hospital based out-patient care the two groups of children differed somewhat, with cystic fibrosis children attending the clinic with much greater frequency. Children who were patients of the Royal Hospital for Sick Children

in Edinburgh tended to be reviewed more frequently than those who were patients elsewhere, but the proportions of each in the two groups were almost equivalent, so that comparability between the groups was maintained.

Although practical inconveniences are more likely to be tolerated when they occur relatively rarely, the problems of transport to and from clinic visits were raised with coeliac mothers to provide a contrast to the C.F. findings. (Section III.B.3). The distances between the homes of the families of each of these 54 children, and the out patient clinic which they attended are given in the next Table (Table V.B.(xiv)).

Table V.B.(xiv). Distance of Family Homes from Clinic

	Coeliac	C.F.
Within walking distance	6%	11%
Across town/city	33%	22%
15 miles	28%	30%
30 miles	20%	26%
30 miles	13%	11%
(In both cases N = homes of 54 children)		

Although the distance between home and clinic was not a factor which distinguished the two groups of families, it was apparent that the particular needs of the C.F. families in respect of transportation to clinic had already been given some special consideration. More than twice as many C.F. children as coeliac children arrived for their clinic appointments by ambulance. Table V.B.(xv).

Table V.B.(xv). Mode of Transport to Clinic

	Coeliac	C.F.
On foot	4%	6%
By private car	31%	37%
By public transport	56%	29%
By ambulance	9%	22%
Other	-	6%
	N = 54 children	N = 54 children

Since coeliac children were only requested to attend clinics, on average, between three and four times per year, they were much less likely than cystic fibrosis children to create financial stresses for their parents by this means. Indeed none of the mothers of coeliac children complained of financial difficulties on these grounds, at the time of the investigation.

The time commitment involved in routine clinic visits had already been examined with reference to the experience of the C.F. parents (Table III.B.(xxi)). The comparative data describing the total time involved, the time spent waiting to see the doctor and the time spent with the doctor are presented in Table V.B.(xvi).

Table V.B.(xvi) The Time Factor in Clinic Visits

(a) Estimates of time spent away from home

	≤ 1 hr	≤ 2 hrs	≤ 3 hrs	≤ 4 hrs	> 4 hrs
Coeliac	-	30%	30%	26%	13%
C.F.	-	15%	52%	11%	22%

(b) Estimates of time spent waiting to see the doctor

	≃15mins	≃30mins	≃45mins	≃ 60mins	>60mins
Coeliac	61%	31%	6%	-	-
C.F.	59%	24%	9%	8%	-

(c) Estimates of time spent with the doctor

	≤ 5mins	≤10mins	≤ 15mins	≤ 20mins	>20mins
Coeliac	17%	48%	33%	--	-
C.F.	7%	28%	48%	13%	4%

N = 54 coeliac families; N = 48 C.F. families

Objectively the C.F. children probably did spend longer in the consulting room than did the coeliac children although estimates of more than 15 minutes at one visit were more likely to come from mothers of two affected children. The main point to be made was that the majority of mothers were satisfied with the period of the doctor's time which was allocated to them and the proportion of mothers who would have liked to spend longer

in the consulting room was the same in each group i.e. 24% of the children's mothers. Once again the expression of this desire bore little relation to the time which those mothers already spent with the doctor on an average visit. (Coeliac, $X^2 = 2.65$, d.f. = 2, $P \simeq 0.30$; C F., $X^2 = 2.23$, d.f. = 2, $P \simeq 0.30$).

Clearly it becomes much harder to achieve continuity of care when children attend the clinic only on a few occasions in the year. It is not surprising then that mothers' responses indicate that only 17% of the coeliac children were seen by the same doctor on each occasion. It is perhaps more surprising that in the matched control group only 13% of all the C.F. children were said to have had any such continuous care. The desire for a more personal relationship between the family and the hospital doctor was expressed by the mothers of 61% of the coeliac and 69% of the C.F. children, all of whom would have preferred their child to have seen the same person at each clinic visit.

In the light of this review of parents' expressed attitudes to three important aspects of their attendance at their child's out-patient clinic, it was interesting then to consider their actual behaviour. The system used to score the regularity of a child's attendance was explained in Section II.B.3. It referred to 'regularity' as a measure of reliability in keeping appointments and as such, it was applicable to both coeliac and fibrocystic patients, in spite of the differences in the frequency with which they were expected to attend the clinic. (Table V.B.(xvii). In these terms the reliability of the families attendance showed and interesting difference when the two groups were compared.

Table V.B.(xvii) Regularity of Children's Clinic Attendance

	Coeliac	C.F.
0. Never/lapsed	4%	1%
1. Rare	2%	19%
2. Fairly Regular	39%	15%
3. Very Regular	55%	65%
	N = 54 children	N = 54 children

There were then, fewer families among the control group who showed poor or sporadic attendance at the clinic, than among the C.F. group but among this latter group a higher proportion of families attended with scrupulous reliability. Some of the factors distinguishing these most reliable attenders from other families were listed in Table III.B.(xxii) Where these factors are not also significant in the pattern of the behaviour of the control group they will not be repeated here. Table V.B.(xviii) lists some factors which seem to distinguish the good clinic-attenders from those whose attendance is less reliable, in the coeliac group. The comparative data from the C.F. families are also quoted.

Table V.B.(xviii) Factors Influencing Regularity of Clinic Attendance.

	Coeliac	C.F.
Mother has a job	$X^2 = 3.20$, d.f.=1, $P < 0.10$	$X^2 = 0.07$, d.f.=1, $P \simeq 0.80$
Having younger children	$X^2 = 4.54$, d.f.=1, $P < 0.05$	$X^2 = 0.21$, d.f.=1, $P \simeq 0.70$
Social class status	$X^2 = 4.31$, d.f.=1, $P < 0.05$	$X^2 = 1.86$, d.f.=1, $P \simeq 0.20$
Mother's anxiety	$X^2 = 9.96$, d.f.=1, $P < 0.01$	$X^2 = 1.22$, d.f.=1, $P \simeq 0.30$
Mother's attitude to clinic	$X^2 = 5.07$, d.f.=1, $P < 0.05$	$X^2 = 0.80$, d.f.=1, $P \simeq 0.50$

It is interesting then that the factors which distinguish families who are reliably good clinic attenders from those who are not so good, are different in the two groups under study. Those who are asked to come to the clinic often i.e. the C.F. group, seem more likely to be deterred by such practical considerations as the distance to be traversed to get there or the cost involved. On the whole the attendance of the coeliac families, who were asked to come less often, tends to depend more on the mother's level of anxiety and her attitude to the clinic. The only practical consideration, to reach significance in its difference between the two factions was the question of younger children who had to be cared for while the mother was at the clinic or brought along with her.

Nevertheless these findings, which were endorsed by mothers' own spontaneous remarks, may be of value to those who are concerned about the regularity with which families attend at clinics. In a potentially dangerous condition like cystic fibrosis then, when frequent appointments are required, some thought needs to be given to the practical problems thus imposed. When the condition is less anxiety-provoking, requiring on the whole less frequent clinic visits then the maintenance of regular attendance on these occasions seems to depend much more on the attitudes of the children's mothers.

Parents' comments upon the conduct of the clinic were summarised by two indices which indicated the emotional tone of their attitudes to the clinic administration and to their relationship with the doctors whom they encountered. The details of the derivation of these indices and of the interpretation of the scale on which they were scored were tabulated in Section III.B.3. The same method was employed to summarise the attitudes of control parents and the corresponding data are presented together in Table V.B.(xix.)

Table V.B.(xix) Tone of Parents' Attitudes to Out-Patient Clinic Experiences

Attitudes to:	Clinic Doctors		Clinic Administration	
	Coeliac	C.F.	Coeliac	C.F.
Critical	4%	9%	4%	8%
Mixed feelings	22%	28%	50%	50%
Highly positive	74%	63%	46%	42%
(Responses from parents of N = 54 children)				

There was understandably evidence of a significant degree of 'spread' in these attitudes and those who held positive attitudes towards the doctors at the clinic were more likely also to express positive attitudes about the clinic (Coeliac, $X^2 = 11.61$, d.f. = 2, $P < 0.01$; C.F. $X^2 = 12.36$, d.f. = 2, $P < 0.01$). However, the personality factors implicated in Section III.B.3. did not seem to be influential in the attitudes of coeliac mothers (Cattell's Q.3, $X^2 = 1.03$, d.f. = 1, $P \triangleq 0.30$; Cattell's Q.4, $X^2 = 0.49$, d.f. = 1, $P \triangleq 0.30$).

In fairness it should be pointed out here that the clinic attended by the majority of these children, at the Royal Hospital for Sick Children in Edinburgh, was a particularly good one both from the administrative point of view and in regard to its staffing, as the majority of parents' responses would indicate. Although among the criticisms expressed there were some valid points made e.g. mothers' desire to be given more information or to be able to see the doctor alone on, occasion, others were inevitably more personal issues, concerning the personality of the parent who expressed them, as much as that of the personnel in question. Although such aspects of parental dissatisfaction were scored in Table V.B.(xix), some personality clashes were considered inevitable, especially in the, at times, highly charged atmosphere surrounding the care of the C.F. children. The only slightly personal point which was made repeatedly by parents was taken note of i.e. the need for a more personal element to be introduced into the system of care of, in particular, C.F. children.

4. Treatment.

The nature of the treatment required to control these two diseases is very different, making direct comparisons of the findings impossible in this part of the chapter. However, the treatment prescribed for the two is similar in the more abstract sense, that in both cases it places a considerable burden, both in terms of practical demands and of moral responsibility, on the child's parents and it is in this sense that we shall attempt to compare the responses of the parents in the two groups.

All but one of the coeliac parents in the sample was still being maintained on glute-free diet. The young woman who had opted out of the dietary regime had been a patient of the Royal Hospital for Sick Children in Glasgow but had since rebelled against inconvenience of it and was, at the time of the study, having a normal diet.

Among coeliac children, dietary restrictions which ruled out such childhood delights as chocolate biscuits, some ice creams and some confectionery, were anticipated to evoke some protest. As we observed among the C.F. children, this protest could either take the form of furtive breaking of the rules of the treatment, or of more flagrant resistance. Once more mothers' assessments of the degree of their children's protests were adopted by this study, on the scale described in Section III.B.4. The relative severity of children's protests against these two treatment regimens are indicated in Table V.B.(xx).

Table V.B.(xx). Children's Protests against Treatment

	Coeliac	C.F.
No protest	33%	17%
Slight protest	31%	31%
Moderate protest	28%	30%
Severe protest	8%	22%
	N = 54	N = 54

Where the mothers were conscientious in their administration of the child's diet protests were notably less likely to occur (X^2 = 7.95, d.f. = 1, P < 0.01). In contrast to cystic fibrosis where particular stages of development seemed to be marked by an increase in the child's protest against treatment (Table III.B. (xxviii)) most of the coeliac children's overt objections to their diet seemed to be concentrated into the preschool years, with the under fives being significantly more likely to register protests against the restrictions than the older children (X^2 = 4.04, d.f. = 1, P < 0.05).

The information which the child had been given to justify the need for these dietary restrictions was considered to be an important point. The explanations given to the children were assessed by the Scheme described in Table III.B.(xxix). An honest explanation of the situation given in an age-appropriate way was considered adequate. However some children had been told "Do as the doctor says or he will keep you in the hospital" or, slightly better, "Do as the doctor says or you will be sick". These examples represent a variety of devices by which children

were given incomplete information in a rather threatening manner. The connection between eating the forbidden foods and illness was established but the element of explanation of the causal link was lacking. None of these children had been purposely misled and only those who were very young had been given no information. This provides an interesting contrast to the explanations of treatment given to many of the C.F. children. (Table V.B.(xxi)).

Table V.B.(xxi) Information Given to Children Re: Treatment

	Coeliac	C.F.
None/Child too young	11%	31%
False/misleading information	-	6%
Incomplete information	13%	28%
Adequate information	76%	35%
	N = 54	N = 54

There was a trend for those who had been given an adequate explanation to be less likely to protest about their diet ($X^2 = 11.01$, d.f. = 6, $P < 0.10$) but it did not reach statistical significance and age was thought to be an important mediating factor in the relationship.

Nevertheless the achievement of satisfactory control over coeliac disease depends to a considerable extent on winning the child's cooperation, to a greater degree than does the control of cystic fibrosis. The question of informing the child about coeliac disease and training his eating habits accordingly, is

then an important one which has to be tackled at as early an age as possible.

By the time of the enquiry 61% of the mothers described their children as being trustworthy in the matter of their diets. Their assessments of the age when their child could first be relied on not to eat gluten-containing foods varied from 1:0 year to 9:0 years with a mean of 4:6 years and a standard deviation of 2.4. 9% of the children in the sample were too young to exercise sufficient control over their environment for the problem yet to arise but 30% of the mothers knew their children could not be relied upon to adhere to the rules imposed without supervision. This seemed to be largely a matter of the age and intelligence of the child. Children over the age of five years were less likely to steal forbidden foods than were younger children ($X^2 = 9.69$, d.f. = 1, $P < 0.01$); more intelligent children were less likely to do so than the less intelligent ($X^2 = 7.24$, d.f. = 1, $P < 0.01$). Not surprisingly then those who had been given an adequate explanation of the situation were also more likely to be reliable in this respect ($X^2 = 44.4$, d.f. = 4, $P < 0.001$).

This is not to say that coeliac mothers then become relieved of their responsibility. They still provide the main meals and their attitude to the importance of strict adherence to gluten-free diet was very influential in the children's attitudes. Table V.B.(xxii).

Table V.B.(xxii) Mothers' Attitudes to Diet

	Coeliac
Strict adherence to Diet	57%
Fairly conscientious, some problems	24%
Some lapses, often intentional	17%
Poor attempt at dietary control	2%
	N = 54 mothers

It was known that in the past the diagnosis of coeliac disease had not always been made with the supporting evidence of the biopsy findings. Since this is the only way to achieve a definitive diagnosis it was necessary to return these children to a normal gluten-containing diet for a time, until the intestinal mucosa has had time to respond, in order to confirm the diagnosis by a biopsy. Some children were then returned to the gluten-free diet, the presence of coeliac disease having been established, and it was hypothesised that such children, having sampled previously forbidden foods, might find it harder to resist their temptation once the diet was reinstated. Some parents had been told that coeliac disease could be outgrown and their children were withdrawn from the diet at the appropriate age. When it was learned that coeliac disease is a chronic enteropathy which, in its untreated form, increases the risk of the development of malignancy in the system, then these children were also returned to the gluten-free diet, after having become accustomed to normal food.

Although 20% of the children in this sample had had a spell

of having a normal diet in 11% of the sample this was at the mother's instigation and in only 9% of cases was it officially recommended as indicated above. The duration of normal diet consumption varied between two extremes; the children had either been off the diet for only a few weeks or for over a year, and within this limited sample it was not possible to distinguish the time off diet among official and unofficial renegades. It was interesting that the 90% of the mothers who admitted seeing changes in their child during this also saw them reversed when the gluten-free diet was reintroduced. Conversely though 11% of the mothers could describe no differences in the child in relation to his diet.

It might have been expected that having required the taste of other foods it would be particularly difficult for the children to stick to the diet thereafter, particularly if no detriment to the child's health was experienced. However within this limited sample no evidence was found to suggest that those who were returned to their diet after a lapse were any more or less reliable than those for whom the restrictions had been consistently imposed ($X^2 = 3.92$, d.f. = 4, $P \triangleq 0.50$). However it must be stressed that this was not a principal focus of our study and may not present a truly representative finding.

Since the provision of food is traditionally 'mother's job' it might be expected that the administration of the coeliac child's treatment would fall even more heavily to the mother's lot than the treatment of cystic fibrosis. It was encouraging then to find that the fathers of as many as 44% of the children in the sample could be relied upon to give them only gluten-free food

should the mother be absent. A further 30% of fathers were willing to try to provide foor for the child, but were more liable to make mistakes. In view of the fact that there were fathers missing from some of these families, this is a very good record indeed, since it leaves only 7 who take no part in and no interest in their child's treatment. 57% of mothers also had at least one other friend or family member who could be reliably entrusted with the child's care if necessary. In a further 26% of cases these people were described as willing but apt to make mistakes, and so only 17% of mothers had no-one to turn to for help in this respect.

56% of the mothers in this group found the organisation of child's treatment particularly time consuming; 33% of them were having some difficulties with the diet at the time of the enquiry and a further 19% had had problems in the past which had since been resolved. Treating a coeliac child, can then, be trying for mothers, and if the problem is compounded by financial difficulties the mother's attitude to the diet may become very negative indeed.

The problem was expected to be diminished for mothers who were able to provide the same meal, or almost the same meal, for all family members, on most occasions. Table V.B.(xxiii).

Table V.B.(xxiii) Frequency with Which Whole Family Has the Same Meal.

Never	4%
Rarely	13%
Often	48%
Always	35%
	N = 54 Coeliac Children

30% of the mothers of coeliac children expressed some resentment of the imposition of the diet. This showed a reflection in their children's attitudes (X^2 = 10.4, d.f. = 3, P < 9.02) making them more likely to protest against the diet, and interestingly in mothers own attitudes to the clinic doctors. Those who were resentful were more likely to express negative attitudes to these doctors (X^2 = 3.73, d.f. = 1, P < 0.10).

In the early stages, and until satisfactory dietary control has been established, coeliac children are commonly prescribed dietary supplements e.g. iron. Although 46% of the mothers described difficulties in the administration of these treatments it should be recalled that they are given at a time when the coeliac child is by definition, below par, and thus more likely to be temperamentally difficult.

Thus, in spite of apparently very different treatment regimens, coeliac disease and cystic fibrosis may present some points for comparison, in the restrictions they impose upon the affected children and in the protests which they thus evoke. For mothers, the burden, especially of the time commitment and the worries, for example of what to tell the child, may usefully be contrasted.

In spite of the additional stresses which we have already pointed out as being active upon C.F. mothers it was interesting that by the time of the study they expressed more confidence in their own judgment in the care of their sick child than did the mothers of the coeliac children. 83% of coeliac mothers, as against 96% of the C.F. mothers felt confident in their handling of that child. Although the operation of defence mechanisms may

be boosting the C.F. mothers confidence we should be alerted by this finding not to ignore any problems which may arise among the control group sinply because they appear to be less dramatic than those presented by the C.F. families.

We turn now to consider the sources of help which have been available to these mothers to aid them in their task.

5. Agents of Help.

Having encouraged the mothers of cystic fibrosis children to express any needs for help which they felt remained unfulfilled by existing services, it was important for the study to create equivalent opportunities for coeliac mothers to request help, in order that the degree of the specificity of these difficulties to C.F. families might be evaluated.

Initially, the same three kinds of needs were explored; for help with the child and his treatment, for help in the home and for help in the identification of reliable babysitters, and it was very clear that mothers of C.F. children were much more likely to need help than were the mothers of the control group. Table V.B.(xxiv).

Table V.B.(xxiv). Needs Expressed by Mothers

	Coeliac	C.F.
For help with child and his treatment	6%	29%
For help in running the home	2%	29%
For help with baby-sitting	-	7%
	N = 54	N = 54

A further 17% of the coeliac mothers did describe some sort of help which they would have welcomed in the past although by the time of the study they no longer had the same requirements, the child being older.

1 mother in the control group and 4 mothers of cystic fibrosis children already had domestic help at the time of the enquiry.

The small numbers of mothers in the control group who had described needs for help, made it difficult to draw any conclusions about the background to these expressed needs. However it was possible to compare mothers' experiences of the effectiveness of Social Service Agencies in meeting their needs.

Once again the record of the Health Visitors was not an impressive one. 46% of the control group and 65% of the C.F. group mothers had had no contact with them and a further 28% of the coeliac mothers and 10% of the C.F. mothers had found the contact that they had positively unhelpful. Although 26% of the control group and 25% of the C.F. group had been helped in some way by the visits of the Health Visistor the results seemed to indicate that this aspect of the Social Services was not equipped to meet the special needs of these mothers.

Among the control group, as among the C.F. sample, there had been a marked absence of other home visitors, with only 3 mothers in the former group and 12 in the latter describing any such occasions. These it transpired were usually visits for the benefit of the caller, in some fact-finding mission, rather than for the benefit of the mother.

20% of coeliac mothers as against 26% of C.F. mothers had had help from a Social Work Department but help from this source seemed to provide relief only for material problems and it began to appear that coeliac mothers would have welcomed some sort of help in their homes, particularly in the early months following the child's diagnosis. Their attitudes to the nature of home visits required are recorded alongside the C.F. findings, in the next Table, Table V.B.(xxv).

Table V.B.(xxv). Requirements of Function of Home Visits

	Coeliac	C.F.
Not in favour of home visits	8%	25%
Available on request for advice with special problems	43%	23%
Available for information and support	6%	25%
Available for advice, information and support	43%	27%
	N = 54 families	N = 48 families

If anything then the enthusiasm of mothers in the control group was greater than that of the C.F. mothers. However their qualifying remarks suggested an important difference. While C.F. mothers had an ongoing need for moral support and for advice about each new problem which developed, the situation of the coeliac mother was a more static one. Although an urgent need for such home visits was expressed in relation to the early months of coping with the sickly child and the subtleties

of his diet, this need was dispelled as the mothers' dietetic competence grew. Thereafter it was noted that the problems were of a much more domestic nature and many of the mothers expressed a desire for occasional meetings with other mothers of coeliac children, perhaps to exchange recipes, to alleviate what could become a rather monotonous diet. Table V.B.(xxvi).

Table V.B.(xxvi). Attitudes of Mothers to Meeting Mothers of Similarly Afflicted Children.

	Coeliac	C.F.
Not in favour	1%	17%
Indifferent	19%	13%
Mildly in favour	24%	23%
Very much in favour	56%	47%
	N = 54	N = 47

Although this point will be raised again in the next chapter, in relation to Parents' Groups, it was of interest to consider here the possible value of allowing coeliac mothers to become agents of help for each other. For the moment, this section will be concluded with a brief consideration of the in-patient aspects of medical care of these children.

6. Hospitalisation

Not surprisingly, the children in the two groups were distinguished by the extent of their experience of hospital. On average the coeliac children had had only half as many

hospital admissions as their fibrocystic contemporaries, spending on average about two-thirds of the amount of time in hospital spent by the C.F. children. Table V.B.(xxvii)

Table V.B.(xxvi) Hospital Experience of Children in the Study

	Coeliac	C.F.
Range of Number of Admissions	1 - 5	0 - 10
Mean Number of Admissions	1.5	2.9
S.D.	1.2	2.4
Range of Duration of Hospitalisation	1 - 25 weeks	0 - 50 weeks
Mean Duration of Hospitalisation	5.5 weeks	9.3 weeks
S.D.	6.2	11.5
	N = 54	N = 54

Only 33% of the coeliac children had been readmitted to hospital at any time since their diagnosis so that the questions of preparation of the children were less relevant. However, all of these mothers said that they had tried to prepare the child for going to hospital and had been able to discuss his experiences with him afterwards. This is a strikingly different situation from that of the cystic fibrosis children. There, these questions were relevant in 32 of the 54 cases, yet in only 18 of these cases had the children been prepared for going to

hospital and in only 21 cases were the procedures discussed with them afterwards, adding more weight to the growing suspicion that cystic fibrosis does indeed have an inhibiting effect on family communications.

Although some C.F. children did exhibit more protest against hospitalisation than did the coeliac children, showing as many as six of the behaviour patterns described in Table III.B.(xxxiii), on average the protest scores obtained by the two groups were the same i.e. mean values equal to 1.3. These protest scores were not significantly influenced by whether or not the child had been prepared for his hospitalisation (Coeliac, $X^2 = 0.52$, d.f. = 1, $P \triangleq 0.50$; C.F., $X^2 = 0.02$, d.f. = 1, $P \triangleq 0.90$) though the protests of children who had been young, under the age of three years at the time of their hospitalisation, were notably greater than those of older children as expected. Furthermore, there was no evidence that children's protests diminished with increasing familiarity with hospital. Children who had been hospitalised on previous occasions were not found to be much less likely to be upset by the experience than were those who had not been hospitalised before. (Coeliac, $X^2 = 3.09$, d.f. = 1, $P < 0.10$; C.F., $X^2 = 3.12$, d.f. = 1, $P < 0.10$).

It was scarcely surprising then that 22 of the C.F. children and 26 of the coeliac children were described as presenting behaviour problems then on their return home from hospital. Those who had protested against their admission to hospital were more likely to be demanding and temperamental on their return home (Coeliac, $X^2 = 14.5$, d.f.=1, $P < 0.001$; C.F., $X^2 = 11.2$, d.f. = 1, $P < 0.001$). However the behaviour of the

coeliac children has to be interpreted with caution since it will be recalled that until adequate dietary control has been established their irritability may be a more direction function of their clinical condition.

Finally then it was of interest to compare the attitudes of parents in the two groups to their children's hospital experiences. The tone of their attitudes was assessed as described in Section III.B.6 and the comparative data are presented in Table V.B.(xxvii)

Table V.B.(xxvii). Parents' Attitudes to their Children's Hospital Experiences.

	Coeliac	C.F.
No comment	-	37%
Positive attitude	69%	43%
Mixed feelings	20%	4%
Negative attitude	11%	16%
	N = 54 children	N = 51 children

Although parents' satisfaction with the management of their child's diagnosis had generalised to their attitude to the child's own experience in the hospital, in the case of C.F. families ($X^2 = 3.49$, d.f. = 1, $P < 0.10$) and a similar trend was described by the control group findings (Coeliac, $X^2 = 3.73$, d.f. = 1, $P < 0.10$). On the other hand, among coeliac parents positive attitudes to hospital doctors, as assessed in Section V.B.3, and positive attitudes to the child's experiences as an

in-patient in their hospital were likely to be significantly associated ($X^2 = 5.73$, d.f. = 1, $P < 0.02$). This was in contrast to the findings among the C.F. group where no evidence of such a relationship was found ($X^2 = 0.93$, d.f. = 1, $p \simeq 0.30$).

The comparative essay of the last two sections has provided now sufficient information about the similarities and differences between the two groups in personal and medical background factors for us now to attempt an assessment of the relative significance of the social and psychological concomitants of each, for affected families.

C. Family Functioning

Three aspects of family functioning were examined in the study of families affected by cystic fibrosis. These were

(1) Financial Pressures

(2) Social Isolation

and (3) Family Relationships.

Each of these aspects of family life is also relevant to the investigation of families affected by coeliac disease.

Although some of the special dietary requirements of coeliac patients are provided by the National Health Service e.g. gluten-free flour and biscuits, the majority of their food is, naturally, paid for directly by their families. Since flour-based foods, which are taboo to the coeliac, tend to be cheaper than those high in protein, it was suspected that some families might experience financial difficultis as a result of trying to maintain their child on a gluten-free diet. It was also suspected that the problem would be worst among families at the lower end of the social scale, whose diet, traditionally, has a higher flour content.

Since many social activities do involve eating it was also anticipated that the dietary restrictions imposed by coeliac disease could be socially inconvenient for the affected person, on occasion. When this person is a child, we may surmise that the difficulties thus aroused could affect all family members.

Again, because eating is a central activity in the family, the fact that one family member requires a special diet creates catering problems, the solution of which may affect family

relationships. For example, if the coeliac child has separate meals, snacks or sweets he may feel left out or his siblings may resent the fact that he has been singled out for apparently, special treatment. If the whole family has the same food, then they must all adopt the gluten-free diet and this involves an element of self-sacrifice which they may not be prepared to make.

Although the findings to be reported in this chapter provide an insight into these three aspects of the impact of coeliac disease on family functioning, they were principally intended to provide comparative data for a better-informed discussion of the concomitants of cystic fibrosis. To this end then, the results from the reduced group of C.F. families will also be quoted in this chapter to provide a ready means of comparison.

1. Financial Pressures

In view of the changes in the status of the national economy during the course of this study, comparisons of actual sums of money quoted by the mothers during their interviews, are unlikely to be illuminating. However, some relative terms may validly be employed. For example, it is still relevant to record that 54% of the mothers of coeliac children described additional expenses incurred by these children while only 35% of the mothers in the other group found that cystic fibrosis had strained their housekeeping budget.

Some of the financial pressures described by mothers of coeliacs were attributable to their difficulties in obtaining

foodstuffs on prescription. At the time of the enquiry, 20% of these mothers were buying flour or biscuits privately because their doctor was unwilling to prescribe enough to meet their needs, and a further 17% of the mothers reported similar difficulties in the past which had been resolved by the time of the study. However, the purchase of special gluten-free foods was not the only economic drain. More than half of these mothers bought some other foods specially for this child, and in 18 of the 31 cases the additional expenditure was estimated as being more than £1 per week.

Clearly the two groups differed in this respect. Only six of the mothers of C.F. children described an increased foodbill as a result of cystic fibrosis. Although 23% of C.F. mothers also paid more for better quality clothing for their C.F. children, an expense which coeliac mothers did not have, the mothers' reports suggested that the financial pressures occasioned at the domestic level, by coeliac disease were greater than those of cystic fibrosis.

Although the financial demands of the two diseases, in these terms, seemed to differ, mothers in the two groups were very similar in their housekeeping needs. Although the mean amount spent, primarily on food, was slightly higher in coeliac households, £11.17 as compared with £10.43 in C.F. households, we have already indicated that inflation is a dominant influence here. However, it is interesting that 63% of mothers in the control group and 55% of C.F. mothers felt that they needed more money and both groups quoted a need for supplements to the value of between £3.00 and £3.50 as being necessary to meet their families needs adequately.

It was again the case in the control group, that the mothers' employment status did not significantly influence her remarks about the financial pressures imposed by her child's disease, although there was trend, not evident in the C.F. group for coeliac mothers who had outside jobs, to be slightly less likely to describe such problems (Coeliac, $X^2 = 5.54$, d.f. = 3, $P \triangleq 0.20$; C.F., $X^2 = 2.15$, d.f. = 3, $P \triangleq 0.50$).

In order that the impact of these chronic diseases on family functioning be fairly assessed, it is important to point out that this factor of whether or not the family is undergoing financial difficulties, is a critical one in the measurement of some other relevant variables Table V.C.(i).

Table V.C.(i) Some Factors which are Influenced by Financial Pressures on the Family.

	Coeliac	C.F.
Mother's ability to cope (as indexed)	$X^2 = 5.01$; d.f.=1, $P < 0.05$	$X^2 = 8.45$; d.f.=1, $P < 0.01$
Mother's anxiety (from Taylor's scale)	$X^2 = 1.84$; d.f.=1, $P \triangleq 0.20$	$X^2 = 5.54$; d.f.=1, $P < 0.02$
Marital stress (from mother's report)	$X^2 = 5.47$, d.f.=1, $P < 0.02$	$X^2 = 9.02$; d.f.=1, $P < 0.01$
	N = 54 mothers	N = 47 mothers

Further information about the families' material assets was once more noted in terms of ownership of household luxuries Table V.C.(ii).

Table V.C.(ii) Ownership of Household Luxuries

	Coeliac	C.F.
Washing Machines	85%	83%
Cars	56%	48%
Telephones	50%	31%
	N = 54 households	N = 48 households

In the control group even more so than in the C.F. group ownership of these commodities is primarily an indicator of social class status, and here the relationship is significant at the 0.1% level. (owns car x S.E.S., $X^2 = 18.9$, d.f. = 1, $P < 0.001$; owns telephone x S.E.S., $X^2 = 15.1$, d.f. = 1, $P < 0.001$).

Although the coeliac families in the control group did seem to have some additional expenditure to meet because of their children's disease, they did have the consolation that it was not a condition which interfered with the family income. In none of these families did parents describe having lost either time at work nor pay, on behalf of their coeliac child. This is in contrast to 11 fathers and 1 mother in the C.F. group who had been absent from work for some reason relating to their C.F. child.

The relative financial standing of the families in the two groups will be reviewed in the Discussion in the next Section. For the moment, the families' social activities, to be considered next, bear not only on the question of social isolation but also on that of their financial status.

2. Social Isolation

The social inconvenience of dietary restrictions was reflected in the responses of 56% of the mothers of coeliac children who described the diet as inhibiting the family's social activities. Only 9% of these mothers however considered it a serious drawback whereas 28% of the mothers of C.F. children found that their families activities had been restricted by that disease. Some instances of these restrictions are tabulated below (Table V.C.(iii)).

Table V.C.(iii). Family Outings and Holidays

	OUTINGS		HOLIDAYS	
	Coeliac	C.F.	Coeliac	C.F.
Family has not had:	48%	39%	37%	59%
Reasons given:				
Cost of illness	-	11%	-	11%
Inconvenience of illness	11%	-	4%	-
Other	37%	28%	33%	41%
	N=54	N=54	N=54	N=54

The results are presented in relation to the restrictions imposed by the condition of each child in the study i.e. 54 in each group. Among coeliac mothers those whose children were under the age of five years were rather more likely to report their activities as having been restricted by the child's condition ($X^2 = 2.94$, d.f. = 1, $P < 0.10$). Perhaps surprisingly, this was

not a significant factor in the C.F. mothers responses (X^2 = 1.33, d.f. = 1, P ≏ 0.30). It will be recalled that mothers' anxiety and their ability to cope with the child and his illness seemed to be influential here in the case of C.F. mothers but this was not found to be so among the control group (Anxiety, X^2 = 0.61, d.f. = 1, P ≏ 0.50; Coping ability, X^2 = 0.13, d.f. = 1, P ≏ 0.70). It was interesting however that there was an association in both groups between the report of such restrictions on family activities and the existence of marital stress (Coeliac, X^2 = 3.81, d.f. = 1, P < 0.10; C.F., X^2 = 19.3, d.f. = 1, P < 0.001).

The notion of social isolation was also reviewed in relation to the more everyday features of the families' activities and mothers were asked in some detail about the regularity of their contacts with family and friends and whether they felt that the child's condition had interfered with their ability to maintain these relationships. Table V.C.(iv). The frequency of these contacts has been simplified from the original five categories of Table II.C.(iv) to three, for clarity.

Table V.C.(iv). Families' Contacts with Relatives and Friends

Frequency of Contact:	FAMILY		FRIENDS	
	Coeliac	C.F.	Coeliac	C.F.
At least weekly	48%	58%	87%	71%
At least monthly	19%	13%	4%	15%
Rarely/Never	33%	29%	9%	14%
Frequency reduced by child's disease:	11%	21%	9%	21%
	N=54	N=48	N=54	N=48

Cystic fibrosis families then, tended to have more frequent contact with their families and rather less frequent contact with their friends than did the coeliac families; they seemed more likely than parents of coeliac children to describe the intrusion of the child's disease into their social activities and were as likely to describe this effect in relation to their contact with their families as with friends.

In contrast to the findings among the C.F. families, (Section III.C.2.) evidence was found in control group of a relationship between the frequency of contact with family and friends and mothers' declarations that the child's disease had been intrusive. Those who saw their family or friends at less than weekly intervals were significantly more likely to feel that opportunities for these encounters had been reduced by coeliac disease (Family, $X^2 = 6.25$, d.f. = 1, $P < 0.02$; Friends, $X^2 = 21.9$, d.f. = 1, $P < 0.001$). Once again social class

differences were found in the frequency of families' contacts with their extended family, and as expected, members of social classes I and II were significantly less likely to maintain frequent contact with their families than were the families lower on the social scale ($X^2 = 12.2$, d.f. = 1, $P < 0.001$).

Coeliac mothers, too, derived emotional support and practical help from these contacts although they were rather less likely to require practical help than were the mothers of C.F. children. Table V.C.(v). Once again, the more frequent the contact the more likely it was to be beneficial to the mother.

Table V.C.(v). Nature of Help Provided by Relatives and Friends.

Relatives	Coeliac	C.F.
Provide emotional support	59%	65%
Provide practical help	57%	67%
Friends		
Provide emotional support	61%	59%
Provide practical help	54%	67%
	N = 54	N = 48

The frequency of mothers' social contacts showed no discernible relationship to their ability to cope (Family, $X^2 = 0.00$, d.f. = 1; friends, $X^2 = 0.07$, d.f. = 1, $P \triangleq 0.80$) although once again more anxious mothers were more likely to describe reduced social contacts, particularly with their relatives as a result of the child's disease ($X^2 = 4.22$, d.f. = 1, $P < 0.05$).

The patterns of social contact then described by the control group mothers differ only slightly from those described by mothers of C.F. children. It will be recalled that in Section III.C.2. an index of the C.F. families social conditions was derived to summarise the findings about the housing, social and financial conditions. The derivation of the six-point scale was outlined in Table III.C.(vi). When this same scale was applied to summarise the findings from the control group index values ranging from 0 to 5 were recorded. The distribution of scores had a mean of 1.8 and a standard deviation of 1.2. The matched group of C.F. families showed a distribution ranging from 0 to 5, with a mean of 1.8 and a standard deviation of 1.1. There was then no statistically significant difference between the families in the two groups in terms of their social conditions.

3. Family Relationships

It was clear from comparisons of mothers' responses from the two groups, that coeliac disease was less likely to intrude in the child's relationships with others than was cystic fibrosis.

Table V.C.(vi). The Influence of The Child's Disease on his Relationships with Others.

	Coeliac	C.F.
None	61%	37%
Slight	20%	15%
Moderate	15%	26%
Marked	4%	22%
	N = 54 children in each group	

These changes in the reactions of others did not seem to reflect the child's age or sex (Table V.C.(vii)) and although there was a trend among the coeliac results for the ordinal position of the child in the family to influence the reactions of others towards him, the trend did not reach statistical significance and was not repeated in the C.F. findings. It will be recalled that coeliac mothers were less likely to experience their child's disease as an obstacle to their contacts with family or friends, so that this question of whether or not these social contacts had been reduced, was not an influential one in the attitudes of these others to the child, as it had been in the C.F. group.

Table V.C.(vii) Factors Thought to Influence the Attitudes of Others to the Sick Child

	Coeliac	C.F.
Age of child (< 5 or ⩾ 5 years)	$X^2 = 0.05$; d.f. = 1, $P \triangleq 0.80$	$X^2 = 1.80$, d.f. = 1, $P \triangleq 0.20$
Sex of child	$X^2 = 1.56$; d.f. = 1, $P \triangleq 0.20$	$X^2 = 0.31$, d.f. = 1, $P \triangleq 0.70$
Ordinal position of child (1st or later-born)	$X^2 = 2.97$; d.f. = 1, $P < 0.10$	$X^2 = 0.56$, d.f. = 1, $P \triangleq 0.50$
Reduced contact, child-family	$X^2 = 3.01$; d.f. = 2, $P \triangleq 0.20$	$X^2 = 8.96$, d.f. = 2, $P < 0.02$
Reduced contact, child-friends	$X^2 = 0.04$, d.f. = 2, $P \triangleq 0.98$	$X^2 = 7.91$, d.f. = 2, $P < 0.02$
Mother's Attitude to child (from Q.252)	$X^2 = 1.88$; d.f. = 1, $P \triangleq 0.20$	$X^2 = 1.92$, d.f. = 1, $P \triangleq 0.20$

There was a suggestion that these significant others might be taking their cue from the attitudes displayed by the child's mothers but again the relationship did not reach statistical significance.

It was interesting that whereas the siblings of the C.F. children were more likely to become resentful of him if family friends or relatives showed him favouritism and yet there was no such link found with the behaviour of siblings in the control group. (Coeliac, $X^2 = 0.02$, d.f. = 1, $P \simeq 0.90$; C.F., $X^2 = 6.00$, d.f. = 1, $P < 0.02$).

A key area in the study of the C.F. families has already been shown to be the broad one of family communications and the findings which emerge seem to suggest that the parents' knowledge of, and response to, the child's prognosis play a key role in the pattern of communications described. This hypothesis may be examined now by comparison with the control group results.

On the basis of the schema outlined in Table III.C.(viii) communications within the families of coeliac children were assessed. The freedom with which parents in the two groups could discuss their child's condition is thus described in the next table (Table V.C.(viii)). The findings from the matched C.F. group are given in parentheses and for clarity the 'nil' category has been dropped from this table.

Table V.C.(viii) Family Communications - 1. Parents of Chronically Ill Children

	Good	Moderate	Poor
Mother-Spouse	76% (49%)	2% (21%)	13% (30%)
Mother-Others	89% (53%)	4% (32%)	7% (8%)
Mother-Child	83% (23%)	4% (45%)	2% (30%)
Father-Spouse	80% (56%)	11% (33%)	9% (11%)
Father-Others	95% (51%)	5% (40%)	- (9%)
Father-Child	77% (24%)	16% (44%)	- (29%)
Mothers : Coeliac Group, N = 54; C.F. Group, N = 47			
Fathers : Coeliac Group, N = 44; C.F. Group, N = 45			

It was evident that the children themselves took their cue in this matter from their parents. Table V.C.(x). Children with coeliac disease clearly felt freer to raise any issues relating to their condition than did their C.F. contemporaries.

Table V.C.(ix) Family Communications - 2. Chronically Ill Children

Mothers' Views	Good	Moderate	Poor	Nil
Child-mother	81% (28%)	4% (40%)	2% (21%)	13% (11%)
Child-others	80% (19%)	5% (45%)	2% (25%)	13% (11%)
Fathers' Views				
Child-father	77% (27%)	14% (35%)	- (23%)	9% (15%)
Child-others	77% (29%)	14% (31%)	- (25%)	9% (15%)
MOTHERS : Coeliac Group, N = 54; C.F. Group, N = 53				
FATHERS : Coeliac Group, N = 44; C.F. Group, N = 51				

On the whole these channels of communication seemed to operate independently, with little evidence of a significant relationship between the levels of communication achieved in separate relationships. A notable exception to this case was that of coeliac fathers. Fathers whose communication with their wives was good were significantly more likely to be able to talk freely to their children ($X^2 = 14.73$, d.f. = 1, $P < 0.001$) and to outsiders ($X^2 = 9.11$, d.f. = 1, $P < 0.001$) about coeliac disease.

Different factors seem to operate to influence the standard of communication reached in each case. For the parents of cystic

fibrosis children these factors were outlined in Section III.C.3. For coeliac mothers social class factors seemed to be highly influential. Mothers in families of social classes I and II were more likely than other mothers in this group to enjoy good communications with their husbands ($X^2 = 5.54$, d.f. = 1, $P < 0.02$) and marital stress was notably more evident among those who did not achieve such satisfactory communications ($X^2 = 33.7$, d.f. = 1, $P < 0.001$). Mothers of coeliac children who had described a more satisfactory management of their child's diagnosis were more able to discuss coeliac disease than those whose experiences at that time had been less satisfactory ($X^2 = 3.88$, d.f. = 1, $P < 0.05$). Not surprisingly then a circular relationship was established between the level of mothers' comprehension of coeliac disease and their willingness to discuss it ($X^2 = 10.5$, d.f. = 1, $P < 0.01$). Although no personality correlates were found to qualify mothers' behaviour, a trend was observed for more anxious mothers to be more likely to seek to discuss the child's condition ($X^2 = 3.21$, d.f. = 1, $P < 0.10$).

Among fathers the situation seemed to hinge on fathers' possession of the relevant information and only the two factors of fathers' level of comprehension of the disease ($X^2 = 8.92$, d.f. = 1, $P < 0.01$) and the adequacy of the management of the child's diagnosis ($X^2 = 5.81$, d.f. = 1, $P < 0.02$) showed any significant relationship to the willingness shown by fathers to discuss this matter. The question of personality factors or of the operation of defence mechanisms did not arise.

These several factors have been explored in some detail since the findings of this study suggest that the openness with which the child's health is discussed within the family is a key area of difference between these two groups, and a source of some of the other divergent attitudes and behaviours later observed.

For the moment this part of the Section may usefully be concluded with a review of parents' own assessments of the impact which their child's disease has had on their family lives.

4. Impact of Child's Disease on Family Functioning

It was interesting that as many as 61% of the control group mothers did feel that to some extent, coeliac disease had modified the pattern of their lives or at least the quality thereof, while only 28% of the C.F. mothers ratified this point. On the other hand, 26% of the fathers of coeliac children, as contrasted with 40% of the fathers of C.F. children suggested that their child's disease had influenced their family lives. While the control group findings seemed to reflect the objective situation reasonably accurately, it may be suggested that the responses of the C.F. group, particularly of the children's mothers may have been modified, in some cases by the operation of their defences.

Table V.C.(x) Family Hardships Occasioned by having a Chronically Ill Child

Mothers' Views	Coeliac	C.F.
No hardship	50%	28%
Less money to spend on the other children	26%	-
Less energy	15%	4%
Less fun in life	5%	25%
Less spare time	4%	25%
Less money to spend on self	-	14%
Less time to play with children	-	2%
Less time alone with husband	-	2%
	(N = 54)	(N = 47)
Mean No. of Hardships Experienced	1.6	2.3
Fathers' Views		
No hardship	66%	51%
Less money to spend on self	11%	11%
Less money to spend on wife	11%	-
Less money to spend on other children	5%	-
Less money to spend on the house	2%	-
Less energy	2%	2%
Less fun in life	2%	27%
Less spare time	-	9%
Less time alone with wife	-	4%
	(N = 44)	(N = 45)
Mean No. of Hardships Experienced	0.3	1.9

Differences were observed in the nature of the hardships described. Coeliac families expressed their difficulties largely in material terms while the impact of cystic fibrosis was described in a more abstract way, with reference to the quality of the family's life in general.

Parents' views of the hardships of having a chronically ill child are compared in Table V.C.(x) following the procedure described in Section III.C.4.

Since, in either event, the brunt of any burden imposed is borne by the children's parents it is relevant to consider in more detail the relative effect of these two diseases on the parents of affected children and it is to this matter that we turn next.

D. Chronic Illness in Childhood - The Effect on Parents

Coeliac disease, like cystic fibrosis, places a high degree of responsibility on parents for the administration of their child's treatment. Their attitudes to this responsibility are important, not only for their ability to provide an adequate standard of care for the child but also for the environment which they create for his personal development. Kershaw (1966) has suggested that every parent of a handicapped child is emotionally vulnerable in everything in relation to that child. Although cystic fibrosis is not handicapping in Kershaw's terms, the findings of Section III.B. seemed to indicate that this vulnerability was not confined to the parental role but that it extended to other aspects of the parents' lives.

In this chapter the findings of the control study will be presented for comparison with the C.F. data in an effort to evaluate more precisely the significance of the effect which cystic fibrosis seems to have on the parents of affected children. It is hoped that as the problems are clarified, so some means of alleviating them may also be suggested.

As before, the views of both parents were gathered and they are presented here to describe the effect of the child's illness on three facets of parents' lives:

(1) The Parent as a Parent of a Chronically Ill Child

(2) The Parent as a Spouse

(3) The Parent as an Individual.

1. The Parent as a Parent of a Chronically Ill Child.

Parents' attitudes to the child's illness and its effect on their lives were explored in a very general way at first. In some respects the lack of visible physical stigmata can be a barrier to parental acceptance of the diagnosis. In cystic fibrosis the child's symptoms may be an effective reminder of this disease but the well-controlled coeliac child probably lacks symptoms to attest to his abnormality, even in the event of occasional dietary indiscretions. It was suspected then, that the incidence of doubt of the diagnosis would be higher among the parents of coeliac children. Mothers of these children were found to be particularly given to expressing doubts, more so than were their fathers, in an interesting reversal of the results of the C.F. Study, where fathers had been found to be more given to doubts than were their wives. Table V.B.(i).

Table V.B.(i). Parents Reactions to their Child's Illness - 1. Doubts.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Doubts the diagnosis	41%	26%	21%	40%
Is willing to experiment with child's treatment	19%	43%	14%	18%
	N = 54	N = 47	N = 44	N = 45

It will be recalled that in the C.F. group practical considerations of the number and severity of the child's symptoms

were influential in mothers' responses, while fathers' responses had a more significantly defensive element in them (Section III.D). This situation too, seemed to be reversed in the control group where mothers' doubts seemed to grow as a function of the time lapse since their child's diagnosis, (X^2 = 3.49, d.f. = 1, $P < 0.10$) and, more notably, of their anxiety (X^2 = 5.46, d.f. = 1, $P < 0.01$). The attitudes of coeliac fathers, on the other hand, seemed more likely to reflect the practical matter of the adequacy of the explanation of the diagnosis which was given in the first place (X^2 = 2.75, d.f. = 1, $P < 0.10$).

The relationship between such doubts and an expressed willingness to experiment with the child's treatment was found to be highly significant in the control sample (X^2 = 15.0, d.f. = 1, $P < 0.001$). It will be recalled that although the C.F. mothers were more willing to experiment with their child's treatment this experimentation rarely took the form of complete withdrawal of treatment and did not seem to be a product of doubt of the diagnosis (X^2 = 0.55, d.f. = 1, $P \triangleq 0.50$).

Parents' perception of the burden imposed upon them by having a chronically ill child was investigated with particular attention being given to the extent to which this burden prevented the parents from fulfilling other family responsibilities as they might have liked. The comparative data on these two points are presented in the next Table - Table V.D.(ii).

Table V.D.(ii). Parents' Reactions to the Child's Illnes -

2. Burden

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Has felt seriously burdened	30%	43%	7%	27%
Has felt unable to fulfil other responsibilities	20%	45%	2%	18%
	N = 54	N=47	N=44	N=45

It was not really surprising that subjective reports from parents should bear out the objective evidence, that the burden imposed on parents by cystic fibrosis is significantly greater than that imposed by coeliac disease. However, the factors which mediate the experience of burden where found to be similar in the two groups. Coeliac mothers with large families to care for ($X^2 = 6.03$, d.f. = 1, $P < 0.02$) and C.F. mothers living in poor social conditions ($X^2 = 6.16$, d.f. = 1, $P < 0.02$) were more likely to feel overburdened. In both groups the burden was lightened if fathers took an active part in the care of their children (Coeliac, $X^2 = 4.73$, d.f. = 1, $P < 0.05$; C.F., $X^2 = 2.90$, d.f. = 1, $P < 0.10$).

In cystic fibrosis, however, the burden which parents have to bear is not only a physical but also an emotional one, for they are required to accept the unacceptable fact that for all their efforts, their child still does not have a normal life expectancy.

Clearly then, these parents must be vulnerable to periods of deep despondency which coeliac parents have little cause to experience. Nevertheless, some mothers of coeliac children did seem to become very discouraged on occasion, by the thought that their child would never be allowed to have certain foods. Although their distress, to them is none the less real than that of the C.F. mothers, it behoves our comparative study to point out the very different levels at which the reported despondency is experienced. Table V.D.(iii).

Table V.D.(iii) Parents' Reactions to their Child's Illness-

3. Despondency and Optimism.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Feels discouraged	24%	70%	-	51%
Feels responsible for child's condition	15%	30%	-	38%
Feels optimistic re: child's progress	100%	77%	100%	82%
	N = 54	N=47	N=44	N=45

It was interesting that even in a disease like coeliac disease, in which there was no clearly defined pattern of genetic transmission, mothers could still be troubled by feelings of guilt or responsibility for any part which, they suppose, some action of theirs may have played in causing the child's problems. The incidence of such feelings in the control group, was considerably

lower than that described among the C.F. mothers.

The seriousness of the mental stress imposed on parents by cystic fibrosis may be appreciated from the finding that almost one quarter of these mothers and one-fifth of the fathers could not allow themselves any glimmer of optimism in their attitudes to the child's future progress. It is in these parents, in whom all hope seems to have been abandoned, that the most grave repercussions of the disease may be expected to occur.

The distress of the parents of C.F. children seemed to be heightened by feelings of isolation, described by 58% of the mothers and 24% of the fathers. The roles of the C.F. Parents Group and of the new home visiting service in tackling these problems were discussed in the previous Section. To some extent it is true that coeliac disease is less likely to have this emotionally alienating effect on affected families, but it should be remembered that the needs of several of these coeliac mothers were already being met, at least to some extent. The opportunity provided by the clinic, for mothers to discuss their domestic difficulties, in relation to their coeliac child, with a woman who had coped with the same difficulties in her own children, was very useful to many of these mothers. Nevertheless, 13% of the control group mothers and 11% of the fathers did describe feelings of isolation as a result of the child's disease. While these feelings seemed to be ongoing among the C.F. mothers, coeliac mothers whose children had been only recently diagnosed seemed more likely to describe such feelings than those who had had longer to grow accustomed to the disease.

Although many of these mothers, like the C.F. mothers, were against "making a hobby of" the child's disease, we have already recorded their eagerness to meet other mothers (Table V.B.(xxvi)). Although only 31% of the mothers in the sample had joined the Coeliac Society this was probably because only 35% of the whole sample had known of its existence. It was interesting that once again, mothers of social classes I and II were much more likely to be members than were other mothers (X^2 = 13.9, d.f. = 1, $P < 0.001$). Even without holding meetings, the Society was able to fulfil an informative function and mothers who were members were more likely to exhibit a higher degree of comprehension of their child's disease (X^2 = 9.84, d.f. = 2, $P < 0.01$). Nevertheless, more than half of these mothers seemed eager to be able to meet others to exchange notes on their domestic management of coeliac disease. Since the potential harm that could be done by such meetings seemed much less than in the case of the C.F. Parents' Group and since there were indications that good could come of it, the study could fine no reasons to oppose such meetings.

When the difficulties imposed by a child's physical condition are compounded with social and emotional problems the source of these problems may not lie in the child himself so much as in the attitudes of others to him and his illness. It has already been shown that cystic fibrosis can bring considerable modifications to parents' attitudes to the upbringing of their child. It was important for the study to establish the significance of these changes and, to do so, corresponding information was gathered about parents' attitudes to the upbringing of children with coeliac disease.

The issue was once more introduced by the general question of whether the parents themselves felt that they had altered their approach to the rearing of the child in question. Their responses are scored in the next Table (Table V.D.(iv)). However it was not sufficient to know how many parents had made concessions, but the extent of their attitude change was also important. In the eight specific issues in which such attitude changes were discussed, parents had had the opportunity of qualifying their responses according to the degree of modification made. Scoring 1 for a slight change, 2 for a moderate change and 3 for a considerable change in their expectations of the child, it was possible to derive an index, taking values 00 to 24, to describe the change in their attitudes. The mean values of this index scored by the two groups are also recorded in Table V.D.(iv).

Table V.D.(iv) The Incidence and Extent of Attitude Change among Parents in Relation to the Upbringing of their Chronically Ill Children

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Incidence of attitude change	56%	41%	28%	49%
Mean score	1.9	4.5	2.5	4.2
S.D.	2.7	4.1	2.3	4.0

Although coeliac mothers tended to say that they felt rather differently towards the child in question the study was unable to

discover many specific instances in which these feelings were translated into child-rearing practice, in many cases. This provided a striking contrast to the responses of C.F. parents. They tended to deny the general suggestion that they might be bringing their C.F. child up in a rather different way from normal, in response to his disease, and yet their responses to more specific questions did indeed indicate that attitude changes had taken place.

Comparisons of the attitudes of parents in the two groups on these specific points are listed in Table V.D.(v).

Table V.D.(v) Specific Instances of Attitude Change among the Parents of Chronically Ill Children.

Less is expected of the child in terms of:	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Personal responsibility	15%	22%	6%	14%
Responsibility for chores	15%	22%	4%	24%
School achievement	15%	31%	4%	27%
Relationship with Peers	9%	27%	4%	28%
Independent decision-making	7%	31%	13%	35%
Activity away from home	9%	36%	21%	45%
Physical activity	2%	41%	2%	39%
	N=54	N=53	N=44	N=51

Figures are given in Table V.D.(v) in terms of the number

of children of whom less was expected, as far as the available numbers of parents would allow. It was clear from these findings that the trend, in practical terms, was as expected, that more concessions were made to cystic fibrosis than to coeliac disease. In view of the small numbers of children in the control group for whom attitudes had been altered, it was difficult to give any reliable estimate of associated factors. As in the C.F. group, the child's age and sex did not seem to influence the parents attitudes. There was evidence however, that more anxious parents were more likely to lower their expectations of the child in question. The more anxious mothers in the control group were more likely to make allowances for the child in terms of his school work ($X^2 = 5.49$, d.f. = 1, $P < 0.02$) while more anxious fathers were more inclined to limit the children's activities away from home ($X^2 = 9.90$, d.f. = 1, $P < 0.01$). In the C.F. group strong associations were established between maternal anxiety and increased caution over the child's physical activities ($X^2 = 4.34$, d.f. = 1, $P < 0.05$) and once again between paternal anxiety and restrictions on the child's freedom of movement outwith his own home ($X^2 = 4.53$, d.f. = 1, $P < 0.05$). It should perhaps be re-emphasised here that, in the terms of these assessments, parents were creating their own reference points of normal child-rearing behaviour and were deciding for themselves whether their upbringing of the chronically ill child conformed to these standards.

One of the most striking features of this analysis was to be seen in the changes wrought in parents' attitudes to discipline in

the light of the child's disease. Table V.D.(vi).

Table V.D.(vi). Proportion of Parents who are less likely to Punish their Chronically Ill Children.

MOTHERS		FATHERS	
Coeliac	C.F.	Coeliac	C.F.
37%	42%	19%	45%
N = 54	N = 53	N = 44	N = 51

C.F. children were notably less likely to be punished for wrongdoing than were normal or coeliac children.

It had already been observed in the C.F. study that trends in parental attitudes were begun when the children were young and then tended to persist until the parents realised, if they ever did, that their lowered expectations were inappropriate. Parents were asked therefore, whether they were happy with their handling of their chronically ill child and whether both parents usually agreed in questions concerning his upbringing. Reports of ambivalence and dissent are recorded in Table V.D.(vii).

Table V.D.(vii). Ambivalence and Dissent between Parents' Attitudes to the Upbringing of their Chronically Ill Children.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Parent has some misgivings re: child's upbringing	18%	40%	-	36%
Parents disagree on child's upbringing	17%	35%	5%	31%
	N=54	N=47	N=44	N=45

It was clear from these findings that the matter of the upbringing of a child with a potentially fatal disease was a particular vexing one for parents, much more likely to create ambivalence in the attitudes of the individual parent and dissent between the parents, than was coeliac disease. We may expect to see the repercussions of these results in the next chapter when the development of the children themselves is considered.

For the present the review of the parents' role may be concluded by a comparison of the participation of fathers in the care of the children in the two groups. Table V.D.(viii). Only clinic visits and the more general aspects of child-rearing were considered here since it was not realistic to expect that the fathers of coeliac children would administer their children's treatment i.e. prepare their meals, regularly, except in abnormal circumstances.

Table V.D.(viii). Fathers' Participation in the Care of the Ill Children.

	Never	Rare	Frequent	Usual
Coeliac fathers attendance at clinic with child	76%	14%	5%	5%
C.F. fathers attendance at clinic with child	38%	38%	8%	16%

	None	Pleasure only	Pleasure & chores
Coeliac fathers' role in child-rearing	7%	25%	68%
C.F. fathers role in child-rearing	20%	20%	60%
(N = 44 Coeliac Fathers; N = 45 C.F. Fathers)			

Although by no means the majority of C.F. fathers took an active part in their child's treatment, they were more likely than fathers of coeliac children to attend the clinic with the children when the opportunity arose. Coeliac fathers, on the other hand, were somewhat more likely to take an active part in their children's upbringing. However, it should be recalled that the mothers of C.F. children were, in some cases, less willing to delegate the care of their children, even to their husbands and some fathers fought shy of the responsibility. By the time of the enquiry however 16 of these C.F. fathers were willing to become more involved in the care of their child while none of the coeliac fathers expressed such feelings.

Thus, although coeliac fathers may appear to be more active in the general aspects of the rearing of their children, the fathers of C.F. children seem on the whole to be more likely to be involved in the medical matters of their treatment and are often willing to take on more responsibility, given the opportunity and encouragement.

We should now move on to consider the effects of these two diseases on parents' marital relationship.

2. The Parent as a Spouse

In view of the genetic implications which distinguished cystic fibrosis from coeliac disease it was not surprising that the former disease should have a greater impact on parents' marital relationships.

Only three of the mothers in the control described any intrusion of the child's condition into their marital relationship. Clearly the number was too small for any meaningful statistical analysis, although it was interesting that these three mothers had a number of characteristics in common. All three were young women, in their twenties, married to manual workers. In each case the indexed adequacy of diagnosis management was low. All three mothers expressed resentment of the burden imposed on them by the child's condition and none of them was coping well with the responsibility. However, in two of these three families, fathers took no part in the children's activities and it seemed highly probable that coeliac disease was only one factor in an already disturbed relationship.

There was no evidence then that coeliac disease had any effect on parents' marital relationships. However, it was still of interest to consider the social activities of the parents as a couple, as an extension of the investigation of social isolation of parents as a concomitant of chronic illness among their children. To simplify the presentation of the comparative data the frequency of parents' leisure outings are considered

only as more or less often than monthly. Since the two values sum to 100% only those going out at less than monthly intervals are tabuled (Table V.D.(ix)).

Once again social class factors were in evidence and among the families of classes I and II in the control group, parents were rather more likely to go out together ($X^2 = 3.26$, d.f. = 1, $P < 0.10$) and mothers were significantly more likely to go out alone, ($X^2 = 4.16$, d.f. = 1, $P < 0.05$) than were those of lower social class. Factors of family size, maternal anxiety or coping ability did not seem to exert a significant influence over the social activities of parents in the control group.

Table V.D.(ix) Estimated Frequency of Parents' Social Outings - Less than Monthly.

	Coeliac	C.F.
Both parents out together:		
Mothers' report	55%	51%
Fathers' report	55%	51%
Mothers out alone	27%	45%
Fathers out alone	-	22%
	N(mothers) = 54	N(mothers) = 47
	N(fathers) = 44	N(fathers) = 45

The findings suggested that the leisure activities of the mothers of C.F. children were most likely to suffer as a result of the child's disease. Cystic fibrosis on the whole then,

seemed a great force for the potential disruption of parents' marital and mothers' social relationships than did coeliac disease. However, to leave the matter here would be to present only one side of the coin. In conclusion of this issue it was necessary to compare the proportions of parents in the two groups who felt that their relationship had been strengthened by their shared experiences in relation to the child's disease.

It was interesting that comparable proportions of parents in both groups endorsed this view. 35% of all the parents in the control group, 43% of C.F. mothers and 33% of C.F. fathers did feel their marital relationship to have been strengthened in this way. No personality correlates of these views could be found but it was clear that mothers who were supported by their husbands in this way were much better able to cope than those who did so alone (Coeliac, $X^2 = 5.06$, d.f. = 1, $P < 0.05$; C.F., $X^2 = 7.47$, d.f. = 1, $P < 0.01$).

One further aspect of parents' lives remains, in which the child's condition may be influential. In the next part of this Chapter the influence of cystic fibrosis and of coeliac disease on the health and welfare of individual parents will be considered.

3. The Parent as an Individual

Once again particular attention was given to the physical and mental health of the mothers, since in both groups the greater part of the responsibility for the child's care fell to their lot. Mothers' responses to be first few questions of their second interview are compared in Table V.D.(x).

Table V.D.(x). Incidence of Health Problems among Mothers of Chronically Ill Children.

	Sufferers		Have sought medical help	
	Coeliac	C.F.	Coeliac	C.F.
Sleeplessness	24%	79%	13%	13%
'Run down/depressed'	61%	85%	37%	52%
'Nervous'	46%	57%	33%	32%
	N = 54	N = 47	N = 54	N = 47

Although the incidence of such complaints tended to be higher among the C.F. mothers it was rather curious that they were little more likely than the control group mothers to seek medical help. When a sum was derived as before (Section III.D.3) to summarise all the mothers' health problems the mean number of complaints recognised was very similar in the two groups (Mean No. of health problems, C.F mothers = 3.0, Coeliac mothers = 2.6). In view of the nature of the treatment prescribed for coeliac children it was interesting that the proportion of mothers complaining of loss of appetite in that group was double that of the C.F. group, 26% as compared with only 13% of C.F. mothers. The interviewer observed that a few of these coeliac mothers seemed to have become almost hypochondriacal about matters concerning their own gastro-intestinal functioning.

However, coeliac mothers were much less likely to see a direct causal link between their own problems and the child's health. Only 17% of them saw coeliac disease as being at the

at the root of their ill health whereas 51% of the C.F. mothers attributed their problems soley to the anxiety provoked in them by cystic fibrosis. A further 31% of the control group mothers and 30% of the C.F. group mothers held their child's condition only partly responsible. Once again it was the most highly anxious mothers in both groups who were most likely to describe such problems (Coeliac, $X^2 = 6.60$, d.f. = 1, $P < 0.02$; C.F., $X^2 = 5.91$, d.f. = 1, $P < 0.02$).

Among fathers troubles of this nature were predictably lower in their incidence than they had been among mothers (Table V.D.(xi)).

Table V.D.(xi) Incidence of Health Problems among Fathers of Chronically Ill Children.

	Sufferers		Have sought medical aid	
	Coeliac	C.F.	Coeliac	C.F.
Sleeplessness	2%	13%	2%	7%
'Rundown/depressed'	11%	42%	2%	9%
'Nervous'	2%	20%	-	11%
	N=44	N=45	N=44	N=45

Nevertheless, the evidence did suggest that the health of the fathers of C.F. children was also affected by their anxiety about their child, to a greater extent than that of coeliac fathers. (Association with father's anxiety, Coeliac, $X^2 = 1.01$, d.f. = 1, $P < 0.50$; C.F., $X^2 = 4.37$, d.f. = 1, $P < 0.05$).

There was evidence too from parents' scores on Taylor's anxiety scale that the parents of fibrocystic children were sustaining rather higher levels of general anxiety than were the parents in the control group. (Table V.D.(xii).

Table V.D.(xii). Parental Anxiety - Scores from Taylor's Manifest Anxiety Scale.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Range of scores	0-37	3-40	0-31	2-30
Mean score	17.6	22.4	11.9	14.8
S.D.	10.3	9.4	7.2	8.6
	N=49	N=45	N=40	N=41

The personal frustrations occasioned for parents by their child's illness were thought to contribute to the stress placed on parents. Coeliac disease did not seem to have the effect of cystic fibrosis in causing fathers to lose time at work to take part in the care of their child but 5 of these fathers did mention that the child's dietary requirements created difficulties for them in terms of geographic mobility but in only one of these cases did this seem to have cost the father a definite opportunity to work abroad which he would otherwise have taken.

Similarly, the problems created by coeliac disease for mothers who wished to work were also less. Only4 of the mothers in the control group as compared with 8 C.F. mothers had described

job problems occased by the till child. 26% Coeliac mothers and 21% C.F. mothers would have liked to have taken outside employment but felt that the child's disease debarred them from doing so. In addition 23% of mothers of coeliacs and 21% of mothers in the C.F. group described domestic difficulties, while 9% of control group mothers and 26% of C.F. mothers experienced personal frustration, as a result of the child's condition.

Several aspects of the influence of the child's disease had been raised and parents were given the opportunity to record not only their experiences but also their feelings about the effect which the disease had had on their lives. The proportion of parents expressing no resentment on any of these counts is recorded in Table V.D.(xiii).

Table V.D.(xiii). Proportions of Parents Expressing No Resentment of Effect of Child's Disease on their Lives.

Coeliac Mothers	C.F. Mothers	Coeliac Fathers	C.F. Fathers
65%	53%	91%	71%
N = 54	N = 47	N = 44	N = 45

Feelings of resentment were clearly more likely to be expressed by the parents of C.F. children than by the control group parents and mothers in both groups were more likely to be resentful than were their husbands.

These feelings of resentment among the mothers were significantly associated with other personal problems described by these women and a vicious spiral of cause and effect can be traced through these relationships.

Mothers who were resentful of their child's disease were more likely to have health problems ($X^2 = 5.06$, d.f. = 1, $P < 0.05$) and to be highly anxious ($X^2 = 12.5$, d.f. = 1, $P < 0.001$) although these relationships did not reach statistical significance among C.F. mothers. Some sequelae of mothers' resentment common to both groups are suggested by the findings of Table V.D.(xiv).

Table V.D.(xiv). Some Sequelae of Maternal Feelings of Resentment in Relation to the Child's Disease

	Coeliac	C.F.
Poorer coping ability	$X^2 = 21.1$, d.f. = 1, $P < 0.001$	$X^2 = 5.02$, d.f. = 1, $P < 0.05$
More negative attitudes held of child	$X^2 = 16.2$, d.f. = 1, $P < 0.001$	$X^2 = 12.3$, d.f. = 1, $P < 0.01$
More problems with the child in infancy	$X^2 = 10.8$, d.f. = 1, $P < 0.01$	$X^2 = 14.0$, d.f. = 1, $P < 0.001$
A more marked response from the child to his illness	$X^2 = 8.6$, d.f. = 1, $P < 0.01$	$X^2 = 6.7$, d.f. = 1, $P < 0.01$
	N = 54 mothers	N = 53 mothers

In view of the damaging consequences of such feelings of resentment it was of interest then to redress the balance of the study by consideration of the question of how well mothers coped with the care of their child. It will be recalled that in Section III.D.3. an index of maternal coping ability was derived. The question of the time spent in treatment by C.F. mothers was replaced in the coeliac group by consideration of the question of the mother's administration of the child's diet. In that group only those whose adherence to the gluten-free diet was most conscientious were awarded an additional point at this juncture. As before then the index of coping ability represented a scale of values 00-18 (See Table III.D.(xiii)).

On this scale coeliac mothers attained total scores which ranged from 5 to 18, showing a mean of 12.8 and a standard deviation of 3.4. This was slightly bettern than the C.F. mothers whose scores ranged from 1 - 16 and showed a mean of 11.4 and a standard deviation of 3.4. However,in view of the increased pressures which are brought to bear on the mothers of C.F. children, it is to their credit that this mean index value is as high as it is.

In contrast to the findings of the original study (Section III.D.3.) it was interesting that the three facets of coping behaviour among the coeliac mothers were all highly inter-correlated i.e. Mothers' ability to care for their children was related to their ability to meet other responsibilities, (Peason's $r = 0.34$, $N = 54$, $P < 0.01$), which was in turn related to their ability to tolerate the situation without undue anxiety (Pearson's $r = 0.49$, $N = 54$, $P < 0.001$). Finally, mothers who

Finally, mothers who were able to care adequately for their coeliac children were more likely also to be able to tolerate their situation (Pearson's $r = 0.49$, $N = 54$, $P < 0.001$).

Since cystic fibrosis, with its life threatening prognosis, arouses fundamental questions about the purpose of life and the nature of death in the parents of afflicted children, in a way that coeliac disease has no cause to do, it was not surprising then to find that none of the parents in the control group felt that their religious beliefs had been shaken by their child's disease. It may be recalled that one in five of the C.F. parents had called these beliefs into question. Although 13% of the mothers in the control group felt that their religion helped them to cope this was rather fewer than the 23% of C.F. mothers who expressed these views.

The influence of the chronic disease of a child on the lives of his parents has been described in this chapter. Several of the findings reported here have important implications for the development of the child and in the next Chapter the same comparative method is used to assess more accurately the impact of chronic illness on the development of the children studied.

E. Chronic Illness in Childhood - The Influence on the Development of Affected Children.

Children with chronic physical illnesses are thought to be more likely to experience psychological and social difficulties than their healthy peers. For cystic fibrosis children, there is another factor, that of the life-threatening nature of their illness, which is also thought to have a disturbing effect on the children's personal development.

Having described a group of children suffering from cystic fibrosis (Section III.E.) it is necessary for us now to compare these findings with those from the matched control group in order to ascertain the extent to which the cystic fibrosis children have special problems and hence, the extent to which they or their families may need special help.

The aspects of the development of these children to be discussed are, as before:

(1) Infant development

(2) The Children and their Illness

(3) The Education of Older Children

(4) The Social and Emotional Development of Older Children.

1. Infant Development (< 5:0 years)

Once again, a brief ante-natal history was recorded from these mothers with a view to establishing the degree of stress to which they had been subjected while carrying the child in question.

This stress was indexed by means of the scale shown in Table III.E.(i), and the mothers of these coeliac children had experienced stresses which ranged from 0 to 5 and showed a mean of 2.0 and a standard deviation of 1.1 over these 54 pregnancies. This was/was not significantly different from the scores obtained by the C.F. mothers ($X^2 = 0.04$, d.f. = 1, $P \simeq 0.90$). Once again no relationship was established between the severity of such stresses during pregnancy and the incidence of early feeding problems in the infant ($X^2 = 0.10$, d.f. = 1, $P \simeq 0.80$).

Since our concern in this chapter is not to reiterate the pattern of events leading up to the diagnosis, but to describe the child's subsequent development, we pause here only to observe that the coeliac children also passed the milestones of motor and verbal development at appropriate ages. Prediagnostic ill-health and hospitalisation could intervene to delay temporarily some aspects of the development of some of the children, but coeliac disease per se could not be said to alter these aspects of the children's early development.

As before, information was gathered about the infant development of all the children in the sample, from their mothers, and particular note was taken of the incidence of certain of the customary childhood problems. The comparative data are documented in Table V.E.(i).

Table V.E.(i). The Incidence of Childhood Problems

	Coeliac	C.F.
Feeding problems	9%	41%
Toilet training problems	19%	67%
Behaviour problems	32%	57%
	N = 54	N = 54

In view of the nature of the treatment which is prescribed for coeliac children it was suspected that they might have had a rather high incidence of feeding problems, even post-diagnostically. In fact, only 5 of the 54 coeliac children were described as poor and finicky eaters at this stage, in striking contrast to their C.F. counterparts among whom such feeding problems were much more commonly described.

Although coeliac infants may also experience loose stools, more often than other children, especially if their diet is not well controlled, and although there has been some suggestion in the past of a higher incidence of bed-wetting among coeliacs than among normal children, toilet training problems were, in fact, less frequently described by the mothers of coeliacs in this sample.

A relationship between undiagnosed coeliac disease and temperamental difficulties has already been commented on, at the beginning of this Section. Since it takes time for mothers to master the diet it was expected that the coeliac infants would show a relatively high incidence of behaviour problems. This was not found to be the case. Problems of attention-seeking and

demanding behaviour were described in both groups but showed a higher incidence among the C.F. infants.

C.F. infants were also more likely to present problems in relation to bed-time and sleep. 58% of the C.F. children had presented problems of this nature during infancy whereas only 13% of the control children had done so.

Among those who were infants at the time of the enquiry the number of difficulties presented by each of the children was summed e.g. children presenting feeding and toilet training problems, but no others, score 2. Difficulties were scored in feeding, toilet training, general behaviour, sleep, fearfulness, happiness and interaction with others. The categories were not all equally relevant to all the infants but they did apply to equal numbers of children in each group. The mean scores for infant difficulties are then comparable between the two groups. The mean number of difficulties presented by the 18 C.F. infants was 3.2 (S.D. = 1.8) while the mean score obtained by the infants in the control group was only 1.4 (S.D. = 1.3). In view of the additional anxiety of the C.F. mothers at this time, described in the previous chapter, it is perhaps not surprising that this should be the case.

The infants in the control group showed slightly higher scores on the Vineland Social Maturity Scale, raning from 90-125 and showing a mean of 107 and a standard deviation of 8.4. This was not significantly different from the scores of the C.F. infants on this scale, which ranged from 75 to 138 and showed a mean of 104 and a standard deviation of 10.6. However, the scores of the coeliac children did not highlight any category of

particular weakness, suggesting that the poor socialisation scores of C.F. infants are describing a real phenomenon and are not simply an artefact of the scale used.

The findings of the study, limited though they were in respect of infant behaviour, seemed to indicate a higher incidence of problems among C.F. infants, which seemed to be closely interrelated to the problems of their parents.

2. The Children and their Illness.

The information which the child has about his condition is clearly an important factor in the interpretation which he puts upon events which befall him, and hence his reaction to them.

Mothers in the control group were asked to assess how much information they thought their child to have about coeliac disease. From their responses the child's knowledge was graded as before (Section III.E.2). If the child had some age-appropriate notion of the effect of the disease on his body and was aware that he would always have this problem and would always require a gluten-free diet, then his knowledge was graded 'adequate'. To have more information than this was considered 'good' and having less was 'poor'. Once again those who were considered too young for the question to be relevant were scored 'nil'.

In this way the extent of the knowledge of the children in the two groups, about their disease, could be compared. The information which the children are thought by their mothers to have and which they themselves showed the interviewer they had, are compared in Tables V.E.(ii) and V.E.(iii).

Table V.E.(ii). Assessments of Children's Knowledge of their Disease.

	MOTHERS		INVESTIGATOR	
	Coeliac	C.F.	Coeliac	C.F.
Good	52%	25%	44%	25%
Adequate	26%	19%	19%	17%
Poor	6%	19%	31%	28%
Nil/too young	16%	37%	6%	30%
	N = 54	N = 54	N = 36	N = 36

The questions of knowledge of the inheritance or of the prognosis were not relevant to coeliac children so that only the aspects of information common to the two groups are considered in Table V.E.(iii).

Table V.E.(iii). The Information which the Children have Re: their Illness.

Child knows:	MOTHERS		INVESTIGATOR	
	Coeliac	C.F.	Coeliac	C.F.
- the name of his illness?	35%	30%	33%	31%
- that he was born with it?	61%	31%	not	asked
- how it affects his body?	78%	46%	78%	47%
- that he will always have the disease and need treatment for it?	52%	39%	64%	56%
	N = 54	N = 54	N = 36	N = 36

The relationship between age and information possessed was a much simpler one in the control group than among the C.F. children. That older children were more likely to know more was shown by analysis of the information which children younger than, and older than, 8.0 years had, in terms of each of the above four facts (Table V.E.(iii)). In each case the association between being older than 8:0 years and possessing that knowledge was significant at, at least the 2% level.

This greater knowledge of the coeliac children testifies to the better communications within their families particularly since the additional sources of information available to many of the C.F. children are less relevant in the lives of coeliac children e.g. hospital wards. The relationship between the adequacy of the

children's knowledge and the extent of their communications with their mothers was significant at the 1% level ($X^2 = 7.60$, d.f. = 1).

Although the control children seem to lack more information than do their C.F. contemporaries the content of that information is objectively, less likely to be disturbing to them than is the knowledge of cystic fibrosis. It is interesting then to contrast the accounts which the parents' have provided of their children's reactions to their condition.

Table V.E.(iv) Parents' Views of Children's Reactions to their Illness

	MOTHERS		FATHERS	
Child is:	Coeliac	C.F.	Coeliac	C.F.
Alarmed?	6%	17%	7%	6%
Pleased by special attention?	50%	36%	30%	35%
A hypochondriac?	19%	36%	5%	41%
Self-conscious?	24%	42%	18%	18%
Frightened?	-	4%	-	6%
Resentful?	39%	26%	36%	22%
Sad?	22%	15%	20%	10%
Worried about his size or shape?	20%	28%	12%	16%
Given to periods of worry about himself?	4%	11%	2%	14%
Liable to show an exacerbation of symptoms if upset?	-	21%	-	10%
	N=54	N=53	N=44	N=51

Although Table V.E.(iv) indicates the range and incidence of the children's responses, it gives no indication of their duration or severity. These two factors were combined in a single assessment, on the basis of which each child's reaction to his illness was graded as being marked or severe, moderate, slight or nil. On this basis the severity of disturbance observed in the children's behaviour in response to the two illnesses could be compared. (Table V.E.(v)).

Table V.E.(v). Severity of Children's Reactions to their Illness - Estimates based on Parents' Observations.

	MOTHERS		FATHERS	
	Coeliac	C.F.	Coeliac	C.F.
Severe	19%	24%	14%	19%
Moderately Severe	19%	24%	11%	23%
Slight	41%	24%	36%	23%
No reaction observed	21%	28%	39%	35%
	N=54	N=53	N=44	N=51

Two major factors seemed to be associated with reactions of this kind among coeliac children. Firstly, the age of the child was an important consideration. While it will be recalled that overt protests were more commonly described among the younger coeliac children, these more subtle reactions were more likely to occur among children over the age of 5:0 years ($X^2 = 15.9$, d.f. = 1, $P < 0.001$). Such reactions were also more likely to be observed

among children who had been off the diet for some time than among those who had continued with it since their diagnosis ($X^2 = 4.16$, d.f. = 1, $P < 0.05$).

In contrast to the C.F. children, the coeliac children's reactions did not seem to be influenced by their knowledge of their disease (Coeliac, $X^2 = 0.82$, d.f. = 1, $P \simeq 0.30$; C.F., $X^2 = 5.49$, d.f. = 1, $P < 0.02$). There was no evidence that always having the same meal as others in the family in any way diminished the reactions shown by the coeliac children ($X^2 = 1.38$, d.f. = 1, $P \simeq 0.20$) but there was a trend for those who reacted less sharply to the dietary restrictions to be more likely to adhere to them ($X^2 = 2.84$, d.f. = 1, $P < 0.10$).

Although the factors associated with the children's reactions show some differences between the two groups the elements of the situation to which the children are responding are, at least in the early years, strikingly similar.

The young coeliac children, like the C.F. infants are more likely to be described as being pleased with the special attention their condition brings to them. As the children grow older and go to school and as the peer group increases in importance so then the awareness of being different is an important precursor of the children's reactions. Since coeliac disease lacks the sinister implications of cystic fibrosis the older children in the control group are spared the deeper anxieties of their C.F. counterparts.

In order to provide a better evaluation of the personal development of these older fibrocystic children it was important to consider also parallel information about the development of the children in the control group. The first phase of this

comparative study is concerned with the children's school performance.

3. The Education of Older Children. ($\geqslant$ 5:0 years)

Controls were found for all of the children described in Section III.E.3 so that only those results which refer to the coeliac children will be reported in full here, to avoid repetition.

The thirty-three coeliac children were also tested individually in their homes on the Wechsler Intelligence Scale. They attained scores which ranged from 83 to 129 and showed a mean of 106 and a standard deviation of No significant difference was found between the intelligence quotients obtained by children in the two groups. ($t = 0.39$, $N = 33$, not statistically significant). To provide a comparison of the C.F. findings, the control children's verbal and performance scores were separated and their relative values are indicated in Table V.E.(vi).

Table V.E.(vi). The Verbal and Performance Scores of C.F. Children and their Controls - from W.I.S.C.

	GIRLS		BOYS	
	Coeliac	C.F.	Coeliac	C.F.
$\geqslant$10 pts. difference, V > P	27%	20%	22%	11%
V = P ± 9	53%	47%	50%	33%
$\geqslant$10 pts. difference, V < P	20%	33%	28%	56%
	N=15	N=15	N=18	N=18

The hypothesis that the C.F. children would be more than usually verbal was not, then, borne out by their W.I.S.C. scores. In fact, there was a counter-tendency in many cases for their scores to be better in performance rather than verbal tests.

All of the coeliac children attended normal schools. Their levels of educational attainment were assessed in basic arithmetic, reading and spelling, again by means of standardised tests administered in the child's home. Mean retardation scores were derived as before (Section III.E.3) to provide some guide as to the extent to which the C.F. children were underfunctioning, when their performance was measured by the same instruments (Table V.E.(vii).

Table V.E.(vii). Mean Retardation Scores of School Children (expressed in months)

	MEAN RETARDATION		S.D.	
	Coeliac	C.F.	Coeliac	C.F.
Arithmetic	9.4	13.1	9.5	11.3
Reading	7.1	9.3	7.1	11.1
Spelling	8.5	11.4	8.8	13.0
	N=33	N=33	N=33	N=33

The mean retardation shown by the C.F. group over all three subjects was 11.3 months (S.D. = 11.8) while that shown by the control group was 8.4 months (S.D. = 8.5). In none of these cases did the difference between the two groups reach statistical significance although the differences in their arithmetic score (t=1.56, N=33)

and between their overall retardation scores ($t = 1.26$, $N = 33$) showed a trend for the coeliac children to perform rather better than those in the C.F. group.

The numbers of children whose performance was poor in more than one of the tests was noted and the proportion of boys and girls in this category is given, with their mean score, in Table V.E.(viii).

Table V.E.(viii) Indications of Underfunction among Chronically Ill School Children.

	GIRLS		BOYS	
	Coeliac	C.F	Coeliac	C.F.
Underachieving by $\geq$ 12 months in >1 school subject	20%	28%	33%	44%
Mean retardation over all 3 subjects (in months)	7m	9m	8m	13m
	N=15	N=15	N=18	N=18

Although these differences are not statistically significant it must be recalled that this was a small sample. It was interesting then that in individual cases among the coeliac children there were identifiable circumstances to which the poor school performance could be attributed in that case and this point will be raised again in the discussion.

For the moment, taking the results only at their face value

there is a suggestion that the coeliac children are underfunctioning in academix terms, although to a lesser extent than their C.F. counterparts. The factors contributing to the performance of the C.F. children have already been considered in Section III so that it is of interest to consider here those factors which seem to be related to the performance of the control children.

A trend was observed once again for the more intelligent children to be more likely to be underfunctioning (Table V.E.(ix)).

Table V.E.(ix) Coefficients of Correlation between I.Q. and Degree of Retardation in School Subjects among Coeliac School Children (Pearson's r)

	Pearson's r	Significance level
I.Q. x Retardation in Arithmetic	0.41	0.007
I.Q. x Retardation in Reading	0.18	0.14
I.Q. x Retardation in Spelling	0.35	0.07

By summing the children's scores on Stott's 'unract' and 'ovract', to be discussed in a moment, it was possible to obtain a general assessment of the degree of disturbance shown in the child's school behaviour. Although this practice is not recommended for a serious assessment of maladjustment it was sufficient for the purpose of showing here that children who

show any such signs of emotional upset in the school environment are likely to produce poor school work ($r = 0.33$, $N = 33$, $P < 0.05$).

Factor's of the child's age and social class background did not seem to be significantly related to performance. Parents' attitudes to the children's schooling were indexed as before (Table III.E.(xii)). Although the question of other educational provision being made for coeliac children was irrelevant, parents' attitudes as summarised by the remaining five items did have a significant effect on the children's school performance ($r = 0.37$, $P < 0.01$).

The practical questions which concerned the mothers of coeliac children were rather different from those worrying the C.F. mothers. They were primarily concerned that their children should not have any gluten-containing foods either from the teacher, in school meals or from their peers. They did not have the same anxieties about the child catching colds or about his toilet functions.

Nevertheless, even without such difficulties the coeliac children were almost as likely to present problems in settling in at school, though rather less likely to have continuing problems after that time (Table V.E.(x).

Table V.E.(x). The Incidence of School Problems among Chronically Ill Children.

	GIRLS		BOYS	
	Coeliac	C.F.	Coeliac	C.F.
Problems on starting school	17%	28%	44%	33%
Continuing problems thereafter	6%	17%	39%	39%
	N=18	N=18	N=18	N=18

Only two of the mothers of coeliac children felt that the child's school teacher was giving him extra attention as a result of his condition. Otherwise, no unusual reactions on the part of teachers were described, indeed in several cases the teachers were not even aware that the child had coeliac disease. The situation for the coeliac child at school then was rather more likely to be that of a normal child than was that of the C.F. child although this rather depended on the severity of the C.F. child's symptoms.

The results of the two parts of this study are more directly comparable when we turn to review the social and emotional development of the older children.

4. <u>The Social and Emotional Development of Older Children</u>

The play of the children in the control group was described

by their mothers. Tendencies to social isolation and to excessive timidity were scored for comparison with the C.F. data. Table V.E.(xi).

Table V.E.(xi). The Social Activities of Chronically Ill Children

	Coeliac	C.F.
Tend to play alone	11%	58%
Always wants to play in own home	11%	47%
Cannot stand up for himself	3%	36%
Takes an active part in sport	61%	50%
	N = 36	N = 36

Mothers' descriptions of their children's social behaviour were summarised by a simple index of social development (Table III.E.(xv)). Higher scores on this scale indicated an increasing tendency to social isolation. The Control children obtained scores which ranged from 0 to 2 with a mean of 0.3 and a standard deviation of 0.6, notably lower than the degree of social isolation indicated by the C.F. group mean of 1.8 ($t = 5.81$, $N = 36$, $P < 0.001$).

Among the control children a significant relationship was found to exist between the degree of the child's response to his illness and the extent of his social isolation ($r = 0.34$, $N = 36$, $P < 0.02$). Among these children, parental expectations, as indexed in Table III.E(xvi) were not an important factor in their social behaviour ($r = 0.0$, $P = 0.50$).

33 of the coeliac children over the age of 5:0 years were still at school at the time of the study. Their class teachers were invited to describe their behaviour at school with the aid of the Bristol Social Adjustment Guide. In each case, a normal child was selected from the same class to meet the criteria given (Section II.D.2g.) and was described, using Stott's Guide, by the same teacher. The distribution of scores obtained by these children and by the C.F. children are given in Table V.E.(xii). The reader is referred to Section III.E.4 for an explanation of Stott's scaling.

Table V.E.(xii). Distribution of Scores obtained by Coeliac Children and their Normal Controls, in comparison with the Scores of C.F. Children, on Stott's Unract and Ovract Scales.

UNRACT	0	1	2	3	4
Coeliacs	22	1	2	5	3
Normal Controls	22	2	2	2	5
C.F. group	21	5	-	1	6
OVRACT					
Coeliacs	15	4	3	3	8
Normal Controls	23	2	3	1	4
C.F. group	19	5	5	3	2
	(N = 33 in each group				

On this basis the incidence of under-reaction described among the C.F. children is not significantly different from that found among coeliac children or their normal controls. On the other hand, there does seem to exist a tendency for coeliac children to be more over-reactive than either the C.F. children or the normal children selected as controls for the coeliacs. This pattern of the results may be amplified with reference to the mean scores obtained by children in these two groups, on these scales (Table V.E.(xiii)).

Table V.E.(xiii) Mean Scores Obtained by C.F. Children compared withthose of Coeliac Children and their Normal Controls, on Stott's scales of 'Unract' and 'Ovract'

Boys	Unract	Ovract
Coeliac	M = 6.0; S.D. = 8.7	M = 14.1; S.D. = 13.8
Control	M = 5.0; S.D. = 10.5	M = 7.9; S.D. = 11.8
C.F.	M = 8.6; S.D. = 15.2	M = 7.9; S.D. = 12.5
N = 18 in each group		
Girls		
Coeliac	M = 3.3; S.D. = 6.9	M = 7.9; S.D. = 10.9
Control	M = 4.4; S.D. = 8.1	M = 3.0; S.D. = 9.0
C.F.	M = 3.5; S.D. = 7.3	M = 3.1; S.D. = 4.9
N = 15 in each group		

Although the trend persists for C.F. children to obtain higher scores for under-reactive behaviour, the difference between these scores and those of the coeliac children does not reach statistical significance ($t = 0.33$). However, the interpretation of these findings depends on more than a head-counting approach and the matter of these children's social adjustment will be raised again in the Discussion.

For the moment it is of interest to note that among the coeliac children boys were more likely than girls to show this under-reactive behaviour ($X^2 = 3.24$, d.f. = 1, $P < 0.10$) and the more intelligent among these children were more likely to be so described (Pearson's $r = 0.31$, $P < 0.01$).

Coeliac children were more likely to be described as showing over-reactive behaviour, particularly the boys in the sample ($X^2 = 2.77$, d.f. = 1, $P < 0.10$). As the response of C.F. children to their illness seemed in several cases to be in terms of under-reactive behaviour so conversely, there was an association between coeliac children's knowledge of their illness and over-reactive behaviour ($r = 0.23$, $P < 0.05$). However, the age of the child and his level of intelligence were also important factors with older children and more intelligent children again being more likely to exhibit these reactions (Age, Pearson's $r = 0.19$, $P < 0.08$; I.Q., Pearson's $r = 0.29$, $P < 0.02$).

It will be recalled that among C.F. children the child's intelligence was also an influential factor in his school behaviour, but so too were the grade of his clinical condition, his anxiety in relation to his symptoms and the extent of family communications about his health. Only the last of these

factors was relevant also to coeliac children, but its influence in their school behaviour was not significant.

28 children in each group were able to complete the Children's Form of the Manifest Anxiety Scale. C.F. children tended to gain higher scores on this scale than did their controls, with mean scores of 18.1 and 15.4 being obtained respectively, by the two groups. This difference between the means did not reach statistical significance ($t = 1.55$, $P \triangleq 0.20$).

It was clear from the findings of this study that the mothers of these coeliac children did not consider that their children had emotional problems of the nature of those described by the C.F. mothers. All of the coeliac mothers considered their children to be of a generally happy disposition. None of these children were described as very fearful and only 4 of the 18 boys and 5 of the 18 girls were described as even moderately fearful.

The children themselves were much more open, on interview, especially in relation to their condition and its treatment. However, there is a danger here that in comparing their responses with those of the C.F. children that the grievances of the coeliac children may fade into significance. In fact, the children who are deeply resentful of their lot feel the bitterness just as deeply as those whose resentment is occasioned by cystic fibrosis. Certainly, equal proportions of the children in the two groups described feelings of being different from others. At the observational level it was interesting that the coeliac children's attitudes tended to parallel those of their mothers more closely

than did those of the C.F. children. Where a mother was distressed by her child's need for a special diet then the child seemed likely to be distressed too.

In fairness though coeliac disease does not pose the same hurdles for these young people. Clearly there was no obstacle to forward planning for these children or their families and the problem of finding suitable employment was for them no harder than for any normal school leaver. Among the eight adolescents in the group two had been bitterly resentful of the social inconvenience of the diet. At the time of the enquiry the oldest girl in the sample (aged 17:8 years) had given up the diet completely. The oldest boy in the sample (13:3 years) opted out only on those occasions when it would have been embarrassing for him not to conform. The remaining six children over the age of 11:0 years had discovered means,which they found acceptable, of maintaining their dietary routine while participating in social activities, with minimal embarrassment.

Before discussing more fully the issues which are beginning to crystallise out from this comparative study it is necessary to complete the picture by giving some thought to the situation of healthy children in the families in these two groups.

F. The Healthy Siblings.

It has already been lamented in Section III.F that the resources of this study did not allow a comprehensive analysis of the responses of healthy children to their fibrocystic siblings. To the impressions already formed of their situation, from that study, we may hope to add some insight, by comparison with the control study of the behaviour of siblings of children withcoeliac disease.

Since the rather special case of the family with two fibrocystic children has already been discussed, such cases were excluded from this analysis, and only those families with one chronically ill child and other healthy children are discussed here.

Although once again, it is impossible to give adequate attention to the factors of the ordinal position, age and sex of the siblings, it is of interest to note the distribution of at least some of these variables in the group as a whole. (Table V.F.(i)).

Table V.F.(i) The Age and Sex Distribution of the Healthy Siblings in the Sample

	Coeliac	C.F.
No. of Families	52	38
Total No. of Well Siblings	111	70
No. of Brothers	62	42
No. of Sisters	49	28
No. of Elder Siblings	77	38
No. of Younger Siblings	34	32

Clearly the numbers of children included in this section shows a gross difference between the two groups but by expressing the data in terms of means and proportions we may hope at least to identify patterns of response from these children in a manner which may allow comparisons to be made.

Since the restrictions upon the food intake of a coeliac child are as apparent to other healthy children in the family as are the treatment requirements of the C.F. child to his siblings, it was hypothesised that comparable needs for explanation of the situation to the siblings would be experienced in both groups. In fact only 12% of the coeliac mothers had not given their other children some explanation of why their sibling needed to have a special diet and in those cases the children were, in any case, too young for such explanations to be necessary. In contrast, 27% of the C.F. mothers had given no explanations of C.F. to their other children. Although, again, in some cases, the children were

too young, this higher incidence of lack of explanation to the other children does reinforce the impressions formed earlier that some families do indeed experience considerable difficulties of communication about cystic fibrosis.

Mothers had also been asked whether they felt that their other children had felt left out as a result of the additional demands placed on their parents, by their siblings. Since objectively, the burden of responsibility imposed upon the C.F. mothers is greater than that placed upon the coeliac mothers, it was not surprising to find that only 25% of mothers in the control group, as compared with 61% of the C.F. mothers, did report that, at times, their other children had felt left out.

The question of whether or not the siblings felt left out when the mother had to attend to the sick child, was an important one, for siblings who were described in this way seemed more likely to present behaviour problems, presumably in a bid to recapture the attention of their parents for themselves. (Coeliac, $X^2 = 3.12$, d.f. = 1, $P < 0.10$; C.F., $X^2 = 5.34$, d.f. = 1, $P < 0.05$). Among the coeliac families the problem seemed at its worst when the children were young, when the siblings were most likely to feel left out ($X^2 = 2.82$, d.f. = 1, $P < 0.10$) and it seemed to be least in the families where fathers took an active part in caring for their children ($X^2 = 12.4$, d.f. = 1, $P < 0.001$). These relationships did not reach statistical significance among the C.F. families. Certainly 55% of the coeliac mothers did report that coeliac disease was an intervening factor in the children's relationships within the family. Although this was a significantly lower proportion than the 74% of C.F. mothers who found that child's

illness intrusive, it still represents an important aspect of the influence of chronic disease in the family. The effect on sibling relationships was investigated more closely, again using the scheme outlined in Table III.F.(ii). For clarity, only the mean scores achieved by elder and younger siblings for each type of reaction are quoted. (Table V.F.(ii)).

Table V.F.(ii) The Responses of Children to their Chronically Ill Sibling

	ELDER SIBS.		YOUNGER SIBS.	
	Coeliac	C.F.	Coeliac	C.F.
Give in more	0.6	2.6	0.0	0.3
Less aggressive	0.6	2.6	0.0	0.2
More protective	1.1	3.4	0.0	0.6
Feel responsible for him	1.1	2.4	0.0	0.4
Jealous	1.2	1.5	0.4	0.7
Worried	0.1	2.4	0.2	0.4
Talk to him about it	0.3	0.7	0.0	0.2
	N=77	N=36	N=34	N=32

The total reaction scores for siblings in the two groups did suggest that cystic fibrosis had a greater impact on sibling relationships, at least from the mothers' point of view, than did coeliac disease. On a scale which ranged from 00-21, coeliac children's siblings showed a mean reaction score of 1.75, considerably less than the mean score of 6.5 obtained by the

siblings of the C.F. children. The meaning of this difference in mean scores is better understood with reference to Table V.F.(ii).

The sisters of coeliac children seemed more likely than their brothers to alter their behaviour in response to the child's disease (Mean reaction score for sisters of coeliacs = 2.1; Mean score for brothers = 1.4). A similar pattern was observed among the siblings of the C.F. children where the mean reaction score of the sisters (5.5) was greater than that of brothers (6.5).

In both groups there was evidence that these children's responses were in part a reflection of the information that had been given to them by their parents. If the mother had explained the situation to the siblings it seemed that she was also likely to have made requests of them in terms of their beheaviour to the sick child, and sibling reaction scores were correspondingly higher (Coeliac, $X^2 = 6.56$, d.f. = 1, $P < 0.02$; C.F., $X^2 = 13.3$, d.f. = 1, $P < 0.001$).

The views of fathers described the same pattern of events, although once more fathers seemed to be less aware than mothers of difficulties among the siblings. 14% of 42 fathers from the control group had noticed an element of resentment in the attitudes of their healthy children to the coeliac children while 26% of 35 fathers in the C.F. group described such resentment.

In view of these findings it was not surprising to find that behaviour problems among siblings were described in proportionately twice as many C.F. families as coeliac families.

31% of C.F. mothers and 15% of coeliac mothers identified such problems among their healthy children.

It was interesting then to discover whether these disturbances extended to the children's behaviour outwith their homes. Some impression of their behaviour at school was obtained by means of teachers' assessments as scored by Stott's Bristol Social Adjustment Guide. As before, teachers were asked to select from the same class, another child of comparable age, sex, intelligence, social and family background, to be described in the same way as the sibling in question. The comparative data for each of the groups of siblings and for their individual control groups are presented in Table V.F.(iii). These data refer only to those siblings who were between the ages of 5:0 and 16:0 years at the time of the enquiry.

Table V.F.(iii) Distributions of Scores on Stott's Unract and Ovract - for Healthy Siblings of Chronically Ill Children and their Controls

SIBLINGS OF COELIACS					
UNRACT	0	1	2	3	4
Siblings	74%	6%	6%	8%	6%
Controls	71%	4%	7%	11%	7%
OVRACT					
Siblings	58%	5%	11%	14%	12%
Controls	78%	10%	2%	5%	5%
SIBLINGS OF C.F. CHILDREN					
UNRACT	0	1	2	3	4
Siblings	56%	15%	5%	9%	15%
Controls	81%	5%	-	5%	9%
OVRACT					
Siblings	55%	10%	5%	15%	15%
Controls	85%	5%	-	10%	-

N = 73 Coeliac Siblings (and Controls) ; N = 34 C.F. Siblings (and Controls)

It may be recalled from Table III.F.(iv) that this

coded scale is interpreted as:-

0 : (near -) stability

1 : mild under/over - reaction

2 : appreciable reaction

3 : maladjusted reaction

4 : severely maladjusted reaction.

The presentation of the data in these two tables is slightly different, results being expressed in percentages here for more ready comparison. These findings are simplified still further in Table V.F.(iv) which shows the numbers of children who obtain scores for at least appreciable over - or under - reaction

Table V.F.(iv) To Show in a Simplified Way the Incidence of Appreciable Reactions among Siblings and their Controls (from Stott)

	COELIAC		C.F.	
	Siblings	Controls	Siblings	Controls
Under-reactive	20%	25%	29%	14%
Over-reactive	37%	12%	35%	10%
	N=73	N=73	N=34	N=34

Since some sex differences are expected in the behaviour pattern of boys and girls at school the results were separated by the sex of the children and the mean scores for brothers and

sisters of the ill children are given with those of their controls, in Table V.F.(vi).

These scores confirm the original impression that over-reactive behaviour is more commonly described among these healthy siblings of sick children than among normal children whose brothers and sisters are also healthy.

Table V.F.(v) Mean Scores Attained by the Healthy Brothers and Sisters of Chronically Ill Children, and by their Controls on Stott's Scales of Unract and Ovract

	UNRACT	OVRACT
Brothers of C.F.'s.	6.2	14.7
Controls	5.3	3.0
(N = 19 in both of the above groups)		
Brothers of Coeliacs	4.2	8.9
Controls	3.8	5.9
(N = 36 in both of the above groups)		
Sisters of C.F.'s.	3.5	4.0
Controls	1.1	1.9
(N = 15 in both of the above groups)		
Sisters of Coeliacs	2.6	9.8
Controls	3.9	1.5
(N = 37 in both of the above groups)		

The findings complete the available information about the chronically ill children and their families who took part in this investigation. These findings and their implications need now to be reviewed in order that the concomitants of the children's illness for these families may be delineated. The discussion of the results which occupies the next Section seeks to achieve this aim.

SECTION VI - DISCUSSION

At its inception, three broad aims were set for this study: Methodological soundness, theoretical interest and practical value. Though they may not be dealt with entirely separately nor, necessarily, in that order, this Section seeks to provide a critical appraisal of the extent to which this investigation has achieved these goals.

The findings of the two parallel studies, presented numerically in the preceding Section, are drawn together here and discussed, to point out the most interesting similarities and differences which have emerged, between the influence of cystic fibrosis and that of coeliac disease, on affected families. By this means we hope to provide the basis for the presentation, in the next Section, of a valid set of conclusions about the social and psychological concomitants of cystic fibrosis, which will be of practical value to those who are concerned with the welfare of these children and their families.

The implications of the similarities and differences observed between the C.F. sample and its control group bear importantly on one of the main theoretical concerns of this study: the evaluation of the role of the prognosis of a child's disease in mediating the psychological impact of that disease on his family. It is hoped that the significance of this factor may be clarified by this Discussion. The other main theoretical issue, that of the importance of the degree of visibility of a disease or handicap, was discussed previously in Section IV and it will be raised again only to the extent that

the findings of Section V cast new light upon the problem.

Throughout this discussion methodological points will be raised as they become relevant.

The Samples

Considerable care was taken in this study in the choice of disease entity from which an adequate control group might be selected. The reports of Section II and V would seem to provide ample vindication of the ultimate choice of coeliac disease for this purpose. However, some methodological difficulties were thereby introduced in the matter of matching the two samples and these difficulties had repercussions for the means by which the resulting data were to be treated. The manner in which these problems were solved is open to discussion, and should be reviewed here, before the findings of the study are themselves discussed.

Clearly there was nothing that could be done about the four fibrocystic children for whom no adequately matched control children could be found. It is encouraging then, to observe that a comparison of the results reported in Sections III and V would not seem to indicate that their exclusion from the control study has substantially biased the report.

However, the matter of the families with two C.F. children had to be considered with a view to action. It will be recalled that the decision was taken to consider each C.F. child as an individual and to assemble a control group then, in such a way as to provide a comparable group of children,

rather than a comparable group of family units. In view of the tremendous individual differences in the manifestation of cystic fibrosis between children and hence, presumably, in some aspects of the concomitants of that disease for them, this decision to consider each child individually was felt to be justified. In some parts of Section V then, these families with two C.F. children were included twice, from the point of view of each of their C.F. children considered individually, thereby providing an equivalent number of responses to that available from the control group.

It was considered that this practice could introduce a serious bias in some aspects of the results, though, so that when the responses of parents were personal and not directly dependent on the condition of the child in question e.g. biographical data, then each parent's responses were scored once only. In that case the total numbers of responses scored in the C.F. sample and in its control group, were not equivalent and the data were expressed as percentages for more ready comparison.

Although this situation was not ideal it was felt to provide the best available solution to the problems presented and attention was then turned to the assessment of differences between the groups in terms of background variables for which it was not possible to control.

Family Settings (Section V.A)

Some important biographical data were gathered from both groups in this study, not only to provide a fuller picture of

the background against which the subsequent findings were to be evaluated, but also to identify poissible sources of error variance which might have been expected to arise from gross uncontrolled differences between the groups on the variables measured.

The backgrounds to the study provided by the families in the two groups were remarkably similar in terms of the factors assessed by this study.

Certainly the parents of the children in the two groups were of comparable age, intellect and educational background. These latter two factors were of particular relevance to the interpretation of findings concerning parents' ability to comprehend information given to them about their child's disease. Some differences were observed between the two groups in the employment status of the children's parents. Among fathers, these differences were interpreted as being of little importance to the study, but the difference between the patterns of employment shown by the mothers of the two groups was of greater interest. 41% of coeliac mothers went out to work while only 26% of the mothers of coeliac children did so. This findings is construed as an effect of the situation under study and, as such, it must be reviewed against the perspective of other effects of childhood disease on parents, but, it may also play a causal role in the determination of other variables under study and, as such, it should also be noted here.

Cystic fibrosis mothers were in many respects very similar to the mothers of the children in the control group but they

were significantly less likely to take up employment outside their own homes and when they did so, it was more likely to involve going out to work as the children were going to bed. Although the numbers of mothers involved are relatively small these differences in the pattern of their employment may be important for later findings.

In a study where so much importance was attached to family functioning, the question of the structure of these families was a central one, although again an area where cause-and-effect relationships were interwoven. From the point of view of background information, the proportion of 'normal families' in each group was established i.e. in which both partners had been married only to each other and had borne children only within that union. The proportion of such families was found to be the same in both groups. The average duration of marriage and the fertility of the families in the two groups, considered as wholes, also showed a high degree of equivalence so that from the point of view of background information about the families, several points of similarity were established. There was a tendency for the relationships between parents in the C.F. sample to be somewhat more volatile than those in the control group. Although the number of broken homes in the two groups did not significantly distinguish the one from the other, the issue of marital stability will, like that of employment of mothers, be raised again later.

In terms of the families' accommodation there was again little significant difference between the C.F. group and its controls. Although the coeliac families were more likely to be

owner-occupiers than were the C.F. families, the living standards of their homes were not significantly different. In fact the coeliac homes were slightly more likely to be overcrowded and in a poorer state of repair than were the homes of the C.F. children.

In addition to the control variables, we have thus established a fairly high degree of similarity between the family settings of the two groups of children in this study. Having satisfied ourselves then, of the adequacy of the control sample on both these counts we are now in a position to review the comparative aspects of the findings of the study.

The Pre-Diagnostic Period (Section V.B.1.)

It has been suggested that the age of the C.F. child at the time of his diagnosis is a critical factor in his parents' response to that diagnosis (Kulczycki et al, 1969). The findings of this study suggest that this is an oversimplification and that it is the events which occur during this prediagnostic period, and not only its duration, which make it a crucial time for these families.

On the whole, there is a wide area of common ground between the prediagnostic experiences described by the mothers of fibrocystic and coeliac children. It was only in a minority of cases of cystic fibrosis that it had been apparent from the child's birth that there was something wrong. In most cases in the study then, the child was at home when his mother first observed that he was unwell. From that time until the ultimate

pronouncement of the definitive diagnosis there was described a period of mounting tension which was highly influential in the mother's emotional state and in her attitude to the medical profession at that time.

The crux of the problem in both cases seems to have been that the diseases in question have not been well known to the general practitioner in the community and that the presenting symptoms were often sufficiently common problems of infancy for referral to specialists to be delayed while the family doctor eliminated the possibility of simple explanations. Problems and delays seemed to have been more marked in cases of cystic fibrosis than in coeliac disease.

Now, many of the doctors in community were trained, before cystic fibrosis or indeed coeliac disease, were understood and the situation may be expected to improve with the passage of time, as those with more recent training take up practices in the community. At present, there is no evidence that the situation has improved in recent years indicating, perhaps, a need for a more active attempt to make these diseases better known to general practitioners.

It was particularly worrying that three social factors seemed to militate against a speedy diagnosis, most commonly in the cases where the child presented as a case of failure to thrive. The young mother, particularly with her first baby was less likely to have her anxieties and fears in relation to her child, taken seriously at first since many young mothers with their normal first-borns do sometimes panic unnecessarily. Mothers of families of lower socio-economic status may have their

child's diagnosis delayed while their family hygiene is queried and food-poisoning or dysentery are ruled out. Rationally such delays are perfectly understandable; the most likely cause of the problem should naturally be eliminated first, but then, during and after these events, more thought should perhaps be given to the feelings of the mother who has from the first feared that her child's condition was serious.

Mothers described a gamut of feelings at this time : feelings of their own inadequacy at their seeming inability to help the child and feelings akin to shame, that their child was not thriving normally. They were often under considerable pressure from friends and relatives who offered well-intentioned advice and suggestions which served only to increase the tension in the situation. When the family doctor was then unconvinced that there was anything seriously wrong with the child it was not surprising that mothers in these cases gave vent to their emotions.

Two main points emerge from the prediagnostic experiences described by the mothers of the children in this study:

(i) There is a need for more information, particularly about cystic fibrosis, to be made available to general practitioners as an aid to earlier diagnosis of that disease

(ii) Given that the events leading up to the referral of the child to the specialist may have been traumatic for parents, greater consideration should be given to the parents' emotional state at this time, both in the management of their child's hospital admission and in the establishment of his diagnosis.

Diagnosis (Section V.B.1.)

As the management of the prediagnostic period is critical to the parents' ongoing relationship with their family doctor, so the management of the diagnosis is important in the establishment of the tone of a new relationship which is to be formed between the family and the paediatrician. Some important features of this management of this communication were discussed in Section IV.

In this Section the diagnosis represents the first issue for discussion in which the factor of the child's prognosis has a crucial influence. From the moment when these parents of the C.F. children in the sample learned that their child's disease was potentially fatal, they were sharply and irrevocably separated in their experience from the families in the control group. A comparison of the findings of the two studies made it clear that the clinicians were already actutely aware of the special needs of the C.F. parents and had given considerable thought to the communication of the child's diagnosis to them. It was hoped that this comparative study would help to clarify some of the issues requiring their further attention.

The purpose of the diagnosis communication in both cases is to explain to parents whatever is the matter with their child, what can be done about it and what the outcome is likely to be. Each of these three aspects is important, especially when, as here, the parents will be required to administer much of the treatment themselves.

In considering the special needs of the C.F. families at this time we should take care not to minimise those of the mothers of coeliacs. There is a danger that in the relief that is felt that the child has coeliac disease and not, for example, cystic fibrosis, that insufficient explanation may be given. Although there was justification for giving this diagnosis to mothers only, if necessary, it was important nevertheless that adequate explanation be given. In several of the coeliac cases in this sample all the information given and sought at this time concerned the administration of the gluten-free diet. While that information was indeed vital, there was evidence to suggest that mothers who did not understand why the diet was necessary or what the consequences were likely to be if it was not carefully maintained, were more likely to succumb to temptations later to break the rules if pressures e.g. financial or the child's protests, were brought to bear upon them. There was a need then, for more complete information about coeliac disease to be given to mothers at the time of their child's diagnosis.

The reverse of this situation seemed to hold in the cases of cystic fibrosis described. Parents became very quickly aware of the outcome of their child's disease and wished to have stressed, rather the role of therapy in its control. However, in fairness to the diagnosing physicians it should be pointed out that in many of these cases a very careful and full explanation was known to have been given. The problem lies in the response of parents to such a devastating communication. A purely rational approach to diagnosis communication is not, then, appropriate.

The emotional responses of C.F. parents to their child's

diagnosis are strikingly similar to the observations made of the reactions of parents anticipating the death of their child from leukaemia (Friedman et al, 1963; Sigler, 1970). In both these instances early reactions of shock allow little information to be taken in and parents' defence mechanisms are quickly mobilised to ward off recognition of the child's danger. Thus, while a considerable amount of information may be successfully transmitted to a single parent at a first communication of a diagnosis of coeliac disease this is unlikely to be a successful tactic in the management of a diagnosis of cystic fibrosis.

It takes time for the awareness of the prognosis to be admitted into parents' consciousness and when it is, it triggers that flood of affects which we have identified as components of the 'anticipatory mourning reaction'. From this study, it would seem that it is only as this "confrontational period" (McCollum and Gibson, 1970) passes, that the parents begin to search for information, as part of their realistic attempt to master the situation and to provide adequate care for the child.

The major point to emerge here then is the different time scale which is required for the communication of a diagnosis with a grim prospect. Whereas the parents of children with diseases such as coeliac disease, may rapidly assimilate the facts of the situation the parents of potentially dying children need to be able to receive the diagnosis at their own pace. Only the most fundamental points need be given at the first confrontation and as we have outlined in Section IV, there will be a need for repeated opportunities then, for these major points

to be reiterated and the details to be fitted in.

Although the management of diagnoses of coeliac disease and cystic fibrosis may differ in terms of their emphases, their emotional tone and their time scale, striking similarities appeared in practical requirements held of them by families in the two cases.

In cases where the child's condition will require treatment to be carried out in the home, as in both these cases, parental understanding of the situation is of the essence. Parents in both groups then welcomed the suggestion that written information might be prepared for them to take home to read after the diagnosis. Even the mothers of coeliac children who are not motivated to forget what they have been told, are, as members of the general public, unlikely to be able to remember all of the information given to them about a complex medical condition at a single telling. In both cases then there was a need for time in which this information could be assimilated before the child was returned home. In this time mothers of C.F. children could be learning about the child's medicines and how to give his physiotherapy, while the mothers of coeliac children might learn to use gluten-free flour and how to plan the child's diet. The need for a continuing source of advice and encouragement to carry mothers through the first months after the child's return home was described by mothers in both groups and this point is taken up again in relation to the agencies of help which are available.

The studies of the psychological sequelae of specific disorders on which Pless and Douglas (1971) base their assumption

that chronicity is the all-important factor do not include any potentially fatal diseases and do not in any case, give any place to the circumstances of the diagnoses of these diseases. The findings of this study would suggest that the influence of a child's disease upon his family begins even before its diagnosis and from that time, that the factor of its prognosis is critical both to the management of the diagnosis and to the parents' response to it.

Medical Supervision - By General Practitioners (Section V.B.3.)

To a layman's eyes the system of medical supervision provided for these children places the family doctor in a rather anomalous position. Having referred the child for specialist attention on account of his chronic disease, he remains nevertheless the doctor who is responsible, nominally at least, for the child's care in the community. The specialist then reviews the child's condition at his out-patient clinic and on the basis of his findings makes recommendations regarding the treatment required, which must then be communicated to the child's general practitioner. This doctor then, usually, adopts the recommendations made and writes the required prescription.

This practice certainly undermines the role of the G.P. for many of these families and when there are delays or confusions in the communication link between the hospital and his surgery then his relationship with the family may become very strained indeed. From the practical point of view this situation can be very tiresome for mothers, particularly of young children, who then have to make not only a journey to the out-patient clinic

but also to the family doctor's surgery and to the local pharmacy before their part of the transaction is complete. It was clear that in some cases mother's frustration and annoyance at this aspect of the system were sometimes given vent at the expense of their G.P.

On the credit side the results of the study suggest that several families in both groups were able to maintain a highly satisfactory relationship with their family doctor. If the doctor knew the family and, if he was perceived by them as being more approachable than the doctors in the hospital, then he was likely to be called upon for emotional support as much as professional advice. This seemed to be particularly true of the lower social class families who were more intimidated by the clinic setting, and of the C.F. families whose needs for support were greater.

The findings of the study seem then to suggest a need for some clearer thinking on the matter of the care of these children in the community. In the case of coeliac disease, where the child once established on his diet may have no special needs, the present system may be satisfactory. However, as control over cystic fibrosis improves these children will spend more and more time in the community and less in the hospital ward so that adequate resources should be available to support them and their families in their home environment.

If the existing system is to be maintained then its wheels need oiling. The communications link between hospital and family doctors needs to be improved in some cases and, in others, more convenient arrangements could be made for mothers to collect

prescriptions. These administrative improvements together with an increase in the knowledge of G.P.'s about these conditions would alleviate some of the problems. The home visiting service discussed in Section IV would, it is hoped, provide a useful addition to this system, by acting as the missing link between home and clinic for these families. In this way the community aspect of medical supervision of cases, particularly of cystic fibrosis could be improved without adding to the already heavy workload of many of these G.P.'s.

Medical Supervision - By Out-Patient Clinics. (Section V.B.3.)

In many respects the system of out-patient care provided for the majority of the children in this sample is excellent and many of the parents acknowledge this. Nevertheless there are still those who cannot be relied upon to keep appointments and there are still minor grumbles even among those who do attend regularly. Some points then may be noted which might make this aspect of their supervision even better still.

The requests made of coeliac mothers in terms of clinic appointments are not heavy. Initially regular visits are made, allowing the mothers opportunities to raise any difficulties which have arisen with the child's diet but once satisfactory control has been established the visits become much less frequent. The majority of these children are fairly reliable attenders although several of these mothers feel that they are bringing the child for the doctor's benefit rather than for their own. Indeed the mothers attitude seems to be a key factor

in the reliability of clinic attendances since those who rarely came to the clinic seemed to hold the attitude that their family doctor could do the same job for them much more conveniently or at least, that the clinic visit represented a wasted afternoon. These views were relatively rare in the group as a whole however.

C.F. mothers in many cases are much reassured by having such regular clinic appointments since several of them live in fear of the child's condition deteriorating suddenly. Nevertheless it must be acknowledged that such regular journeys to the hospital can create stresses of one sort or another for these families. The issue of mode of transport used has begun to be tackled but other factors, e.g. the time commitment, may be very difficult to accommodate. Given that these regular appointments are medically not only justifiable but vital to the maintenance of a high standard of care for the children and that some sacrifices on the part of the family in terms of time, energy and money may be inevitable if these appointments are to be kept then it seems there is a great obligation to make these clinic visits rewarding experiences for these families.

The principal reward sought by families in both these groups was a higher degree of feedback about the condition of the child for this was a much needed source of reinforcement for their efforts in home treatment. Since the other matters concerning clinic supervision, e.g. opportunities for continuity of care or for private consultation with the doctor referred primarily to the C.F. families and were thus discussed in Section IV, we may now pursue further this question of the demands placed on the family in the treatment of the child.

Treatment

Although as Debuskey (1970) points out, there is great consolation to be gained from the fact that these are treatable conditions and amenable to treatment which can be administered in the family setting, the situation is not without its problems.

Treatment of such chronic diseases in which there is no reward for effort in terms of the child being cured, or even necessarily, in terms of visible improvement, can be very emotionally draining. How much more so then, when the child may not only not improve, but may actually get worse, in spite of parents' efforts.

Previous studies of cystic fibrosis e.g. Pinkerton (1969), have stressed repeatedly the importance of maintaining a positive attitude to therapy in the parents of these children. To a lesser extent the same may be said of coeliac disease. The findings from the control study seem to suggest that the control of coeliac disease can however be made more satisfying for mothers than can that of cystic fibrosis, however. Mothers who see the dietary regimen as a challenge are rewarded by seeing their child being otherwise quote normal. Even they may become discouraged, though usually when other problems are also present, and may require some encouragement.

The question of reinforcement, encouragement or support for parents committed to a lifelong routine of treatment for their child is an important part of the philosophy of management, which is required to sustain them through their task.

Discussions with these parents suggested some needs common

to both groups, but which predictably take on a deeper significance for those for whom this is, literally, a matter of life and death. In the first instance there is a need for parents to achieve competence in giving the required treatment. They need to understand what they are doing, why they are doing it and the consequences if they do not do it. Understanding of treatment procedures, especially of so many and complex aspects as may be required by the C.F. child, will take time and patient explanation on the part of doctors, to achieve. Nevertheless the findings of this study suggest that this is an important step in winning the co-operation of parents.

Several mothers who had difficulties in administering some treatment to their child expressed the view that the doctors did not fully appreciate the domestic problems created by their requests. This was less true of the coeliac mothers who had another mother available at the clinic to whom they could refer, but it did apply for instance to the duration of physiotherapy requested for C.F. children. On the whole then a need emerged for those who supervised the child's treatment to provide some advice for mothers about its domestic administration, not only in practical terms, but also if necessary in terms of explaining the situation to the child in such a way as to win his co-operation. This point was allied to the important need for parents to feel part of the team who were caring for their child and fighting his disease. Some parents clearly felt theirs to be a solitary role, the performance of which was likely to be criticised by 'the experts'. This is clearly an attitude

to be discouraged.

Finally, then, parents who are involved in an arduous daily routine of treatment for their child need to be encouraged for their efforts, perhaps to a greater extent than they have been in the past. The only real source of such positive reinforcement which is available to them, in a controlled way, is the approval of those others who are also concerned with the child's welfare. It seems that by incorporating parents into the therapeutic team in this way their burden may be shared and become more tolerable.

Agents of Help. (Section V.B.5.)

Clinic visits however, are fairly short and, at least in the case of coeliac children, rather infrequent, so that the schedule of reinforcement which they provide may not be enough. This is particularly true in the early months following the child's diagnosis when the mothers in both these groups found the existing agents of help inadequate to meet their needs for advice and reassurance.

Whereas the mothers of coeliac children seemed to require home visits only on a few occasions till they became familiar with the diet, the need of the C.F. mothers was an ongoing one. Although these mothers were not, on the whole, overtly preoccupied with thoughts of the child's death it was certainly the case that the prognosis once uttered, was never forgotten and was never far beneath the surface. Thus although the long term needs of these mothers included information and practical advice about any eventuality which might arise, the key issue was clearly one of providing moral support.

This need has been recognised by other authors e.g. Pinkerton (1969), and it seems to be a need which is not equally felt by all parents of children with chronic illnesses but which is particularly acute among parents who have a child with a potentially fatal disease.

The Social Scientist and his Study

One possible source of bias in these findings needs to be acknowledged here, that of the influence of the presence of the investigator in the situation which he is studying. It is entirely possible that the presence of a psychologist in the clinic, concerned with the attitudes of the parents could have wrought changes in some of the factors under study, which would have been effective to differing degrees at different periods during the study and hence would have introduced a bias into these findings. Although the introduction of such uncontrolled variance was to be regretted, it was to some extent inevitable for it added a great deal to parents' approval of this study that it was carried out under the supervision of their paediatrician. This link between the study and the clinic was maintained by the author's attendance at clinics. Such bias, if it was created was most likely to influence those findings just discussed rather than the more personal aspects to which we now turn.

Financial Pressures. (Section V.C.1.)

For National Health Service patients neither of these diseases

should, theoretically at least, impose undue financial stress upon parents. Nevertheless each in its ways may create economic problems for parents and where this is the case, help may be needed.

For the parents of C.F. children financial pressures may be imposed by the need for regular clinic or hospital visits. The provision of better food, warmer clothes and more toys may be part of the wider moral issue of parents' attitudes to the upbringing of a potentially dying child but as sources of financial pressure they are not imposed, nor are they always experienced.

For coeliac families the position is slightly different. Costs of transportation to the clinic are relatively low, the question of damage to housing does not occur and the essentials for the child's diet are provided on prescription. Although it is true that the foods which the child could not have tended to be the cheaper items, it was clear that, in some cases, help with budgeting for the diet of the whole family was sorely needed.

An awareness of the financial pressures bearing upon these families is, then, important. There was evidence that the existence of financial stress is singificantly associated with marital stress, poor coping ability and, among coeliac families, with high anxiety. The economic difference between the situation of the families described in this sample and those described by American Writers is probably largely attributable to this difference, then. It seems highly likely that the financial hardships described among families who have to count the cost of medicines and visits to the doctor, are responsible for the greater

incidence of severe psychopathology among these families.

Social Isolation. (Section V.C.2.)

It is rather difficult to obtain a true assessment of the extent of a family's social isolation without knowing what they did before they had their chronically ill child. Nevertheless other authors e.g. Kershaw, (1966) have pointed to the dangers of isolation which exist. Hewett (1970) on the other hand found no significant difference between the patterns of outings among the families of her cerebral-palsied children and normal children.

This study has considered two aspects of social isolation. In the case of contact with family and friends isolation seemed to be at its worst in the prediagnostic stage when the anti-social symptoms of the undiagnosed child were an embarrassment to mothers in both groups. This situation was analogous to that described in families with visibly handicapped children e.g. by Kershaw (1966). This situation usually improved after the diagnosis was established and the symptoms were brought under control. Mothers of very young C.F. children were afraid of their babies catching germs from social contacts; mothers of children with toilet problems were anxious to save their children the embarrassment of having to use strange toilets; mothers whose friends or relatives would encourage the child to have 'just one' forbidden biscuit; these mothers were likely to describe some consequent social isolation. On the whole, neither group presented evidence of major disruption, on a large scale,

to relationships with family and friends.

Problems did arise for these families in outings and holidays. The inconvenience of having to take gluten-free supplies everywhere with them or of having to search for appropriate foods on a commercial menu was daunting to some families of coeliac children but only a small minority of their families felt their leisure activities to have been restricted as a result. The financial factor seemed to be more of a deterrent to C.F. families although there was an undertone of anxiety lest the child become ill in a strange place. Those who did have holidays tended, like McMichael's sample (1971) to make self-catering arrangements or to have caravans.

The isolation which chiefly concerned this study was not social isolation but the other important
made by Turk (1964) of family communications. Disturbances in communications seemed capable of producing a much more fundamental and more impenetrable kind of isolation than the lack of social encounters could.

Communications. (Section V.C.3.)

Two principal points require to be made about family communications in the light of the results of this study. The first concerned parents' feelings about the child and his illness and the second concerns the child's prognosis as an inhibitor of family communications.

In relation to the first of these points,

Younghusband (1970) made the very telling observation that there is some pressure on all parents of handicapped children to be impossibly good, forever understanding, forbearing and self-sacrificing. He pointed out that any feelings of resentment arouse feelings of guilt in these parents lest they be or be thought to be, rejecting their child and he asks whether parents discuss their feelings between themselves or whether they pretend that they do not exist.

This study would seem to suggest that where the marital relationship is strong and stable parents are indeed likely to share their feelings with each other. However a substantial proportion of parents, particularly among the C.F. families had clearly never acknowledged their feelings to anyone. It would be naive to suggest that all of these parents then confided frankly in the investigator and this was not the case. However, on several occasions during the C.F. study mothers did observe that they had not discussed the child's condition in terms of their personal feelings before and that it was a relief to do so. These mothers did not feel that friends, family or even their spouse would understand their feelings and such matters were never discussed. Clearly some feelings are more acceptable than others and it seems quite likely that this study missed some of the deeper feelings which parents could not acknowledge even in the atmosphere created by this interview. It was felt that in some cases, stress on mothers in particular might be alleviated by providing safe opportunities for these mothers to express their feelings. It was felt that an opportunity for this might be built in to the home-visiting service.

The second point emerged from the notable difference in the patterns of communication observed in the two groups of families. Death, in our society, has become an uncomfortable and embarrassing topic to be discussed only when necessary and the development of public and private attitudes to death to this point would provide a thesis topic in itself but, at present death is something which most people prefer not to think about. Among families affected by cystic fibrosis the issue takes on an even higher emotional tone in relation to their child's prognosis. Since such thoughts are painful, the defences are mobilised to reduce the threat. However, by a process of association, other topics become linked with the threat to the child's life and also become taboo. For example, talking of plans for the future reawakens the thought that the child's future is uncertain which is distressing and is threfore to be suppressed. By this process the child's prognosis can wreak havoc among family communications and varying degrees of defensiveness were observed among the C.F. families in this sample. If this system worked satisfactorily there would be no reason at all to intervene, but, in cases where the child is aware of his illness but can find no one in the family willing to discuss it with him, the effect can be devastating. Pinkerton (1969) observed that both in adults and in children, "fear of the unknown represents an important source of anxiety, so that in the absence of reliable information patients and parents alike will tend to fill the vacuum with their private horrific phantasies."

While the findings do support any move to help break down

some of these communication barriers which can create so much private torment for parents and children alike, we have to point out once again the dangers of undue candour in communication with the children. While several of the American paediatricians advocate telling the truth to a dying child when he asks the dreaded question, "am I going to die?", it must be pointed out that the children in this sample are young children with an uncertain prognosis. For many of them then an emphasis on preparation for life rather than for death is much more important and the morale-crushing blow which may be dealt the child by an inappropriate communication of his prognosis may leave a permanent scare. On the other hand, these are children of normal intellect who will, sooner or later ask questions about death as they will about other fundamentals in life and their parents need to be prepared to provide an answer just as they would if the child did not have cystic fibrosis.

Many families in the sample could handle their interpersonal communications within and outwith the family circle quite satisfactorily. Nevertheless this must be acknowledged as a particularly vulnerable area in which help may be required. Families whose chronically ill children have a poor prognosis may be particularly in need of help of this kind.

Parents of Chronically Ill Children. (Section V.D.1.)

Other particularly vulnerable areas which have been recognised from the first are those of parental acceptance of

the child's illness and of their attitudes to the child himself. The use of these umbrealla terms has already been contested (Section I.C) but some of the concepts subsumed under these headings were recognised as being of importance for the study. Here the attitudes of parents to the child's illness is a case in point. Particular note was given here to the situation of parents of coeliac children. In the absence of symptoms and physical stigma it was sometimes hard for these parents to believe that there was anything wrong with their child and although this doubt did not always cause parents to experiment with the child's diet it did seem to increase the likelihood of the experience of the treatment as an undue burden. The attitudes of parents to cystic fibrosis have already been discussed in Section IV, leaving little to be added here.

Turning then to the matter of parents' attitudes to the child in question the findings require a little more discussion. The results of the study as a whole suggest that mothers do tend to feel differently about their chronically ill children in rather subtle ways, which not all of them felt competent to describe. Whether these rather special feelings were translated into action was found to depend on a number of factors including the child-rearing principles of the mother and the nature of the child's condition. Although the author does subscribe to Hewett's view (1970) that 'the point at which reasonable care becomes overprotection' is hard to establish, the motivation behind these attitudes does provide a clue.

Among the coeliac parents a tendency to identify too closely with the child tended to lead them to pity their child and to make concessions to him by way of compensation. Although this

situation was found to exist in only a minority of cases it did seem in those instances to create an unhealthy environment for the personal development of the child.

It was only natural that the desire to protect and indulge their child should be stronger among parents who feared the curtailment of his life span but there was evidence to suggest that undue changes in the attitudes of the C.F. parents could lead to illness-centredness in the child, disagreement between the parents and resentment among the siblings. All of these problems may then act to compound the existing difficulties.

There was no evidence in the study of either group that such attitudes masked underlying rejection of the child, in contrast to the findings of some other studies (Wright,1960) but rather suggesting that the attitudes had developed from the child's infancy under the influence of the threat to his life of the prediagnostic experiences and of the shock of the diagnosis. By the time of the enquiry several mothers were very ambivalent about their upbringing of the child in question. Although the proportion of such mothers who were unhappy about their child-rearing was smaller than that described by the Newsons (1968) it was clear that the ambivalence in this study was occasioned by parents uncertainty of the stand which they wished to take in the rearing of a potentially dying child.

Section IV has already indicated that this study would not presume to tell parents how to bring up their children but the findings of these two studies would suggest that this is an issue where the two factors of chronicity of illness and prognosis are both influential. The evidence suggests that whatever the

child's complaint there is a risk that it will alter significant aspects of the parents' attitudes to his upbringing and that when the prognosis is grim the effect may be expected to be even more pronounced. Now an ill or handicapped child cannot be expected to have the satisfaction of being just like other children and the difference has to be faced realistically, but in this section we seem to have uncovered a potential danger that real potential which the child has may be allowed to develop and that his personality may be distorted by inappropriate concessions being made. These results point to a need for those who care for these chronically ill children to be concerned not only with their clinical condition but also with their personal development, a point which we shall return to again in a moment.

The Parent as Spouse

Among the parents of chronically ill children there is ample room for marital disharmony. Disagreements between parents over the handling of the child were described, particularly if the mother was more inclcined to indulge the child than was her husband; disagreements were recorded over holidays and leisure activities and over the extent of father's participation in the care of the child. Although the child's illness added a new dimension to the issue the topics are not unfamiliar to normal parents and it was not surprising that where the problems which existed were on this level the child's illness was not seen to have affected the parents marriage relationship in a significant way.

However, when two new elements of genetic responsibility for the child's illness and the fear of the death of the child are introduced the situation presents new difficulties which the parents may find it hard to tackle. As counselling facilities for genetic or marital problems are available to other parents so should they be to the parents of chronically ill children. However there is a danger here that in advocating a global approach to the care of the child that we make a complete takeover bid for all the family's affairs and in the eyes of the author this is not seen to be justified. In identifying the concomitants of chronic illness in childhood we have to acknowledge that marital stress may be a problem. We have identified some specific issues in which valuable help may be given e.g. genetic counselling and in the terms of this study such attempts to alleviate the additional pressures upon the relationship seem to describe the limits of the intervention which can be offered by those who care for the child unless marriage guidance counselling is specifically sought by the parents in question.

Parents as Individuals. (Section V.D.3.)

Certainly physical ill health in parents, particularly mothers may be expected to reduce their ability to cope with the demands placed upon them by having a chronically ill child. However the most important aspect of physical ill health, which was relatively rare among parents of this study, was, in this respect, that it made these parents more vulnerable to

feelings of guilt or responsibility for the child's condition. This was found to be the case too with parents who had been ill as children and the parents feelings in this matter were exacerbated by being asked to give their medical history. It seems that even where no connection would be imagined between the illness of parent and child, parents, in their search for meaning in the situation are likely to assume a causal relationship. An awareness of lay ignorance in such medical matters if necessary on the part of doctors in order that such misunderstandings may be satisfactorily explained to anxious parents whose feelings about the situation may inhibit them from asking directly whether any such causal relationship exists.

The most serious matter to emerge in relation to the health of these parents concerned the very high incidence of sub-clinical mental ill health. Unfortunately no corresponding data is available from the Newsons' studies (1963, 1968) to allow us to assess the extent to which this is truly an abnormal feature. Certainly both McMichael (1971) and Hewett (1970) described depression among their samples of mothers but in Hewett's sample this tended to be associated with the mother's general health with mothers in poor health being more given to depression.

In this study coeliac mothers were likely to attribute their state of health to a variety of causes but as we have already observed the child's condition was the principal factor in the health of the C.F. mothers, once again the child's prognosis was seen as a prime agent in this situation. Although these mothers could not have been said to be preoccupied with morbid thoughts as has been suggested e.g. by Lawler et al (1970), the

knowledge of the prognosis and the burden of responsibility placed upon these mothers did place them under considerable nervous strain.

In spite of this strain many of these mothers coped amazingly well; their strength and courage in doing so is to begreatly admired. The study of the influence of the chronic disease of a child naturally tends to focus on mothers. Among the mothers in this investigation the three broad patterns of parental response identified by Hewett (1970) were again to be found. There were some mothers, most commonly coeliac children or of mildly affected fibrocystic children, who did indeed "give the impression of having had little difficulty in being very matter of fact to the child and to the total situation". There were other mothers who had had to draw upon "physical and emotional resources which they did not know they had" to meet the challenge of hacing a child more severely affected by cystic fibrosis, and finally there were indeed in both groups, those "whose resources were already stretched to the limit" and for whom the chronically ill child was "the last straw". It is for this latter group of mothers who most urgently need all the supporting services which we can provide to care for their children.

Parental Personality

It will be recalled that Cattell's 16 P.F. Questionnaire was given to parents to provide the opportunity of identifying personality correlates of particular aspects of parents' attitudes. In fact, such correlates were rarely found and the

results of giving this test rarely appear in either Section III or Section V. There are a number of reasons for this difficulty which should be acknowledged here.

For parents who had already been most tolerant of a cohnsiderable demand made on their time by this study, these questionnaires were rather too long and for many parents, too intimate, in the context of these investigations. Although there is no reason to suppose that those who did complete these questionnaires did so inaccurately it must be admitted that their motivation was not of the best.

The second point lies in the responsibility of the investigator. With a wealth of rich interview data to be analysed the analysis of the 16 P.F. was cumbersome. It was undertaken in a simplified form so that the analysis sought only to establish the significance of any differences which might exist between the attitudes of those who scored high or low in any given Factor. This oversimplification may have done Cattell's work a grave injustice but within the constraints of the available resources such a simplification of the taks was expedient.

We have to acknowledge then, that the 16 P.F. scores did not make a particularly useful contribution to our understanding of the problem in hand, except perhaps, in terms of Facto B of intelligence. Cattell's 16 P.F. was not an appropriate choice of test for inclusion in this study.

Chronically Ill Children.

The influence of cystic fibrosis upon the development of

affected children has already been discussed in Section IV. Since each of the children in this sample, whether coeliac or fibrocystic was seen as an individual in a unique set of circumstances it was very difficult to make generalisations but the function of the discussion of the children in this section was not to reiterate points made previously but rather to examine any new light which might have been thrown on these points by the findings of the control study.

Infants (Section V.E.1.)

It has been suggested, in relation to the study of the psychological concomitants of chronic illness in childhood that the chronicity of the disorder is of greater importance then its clinical attributes (Pless and Douglas, 1971). Although in its broadest sense the author would agree that there tend to be more similarities than differences between the findings of studies of specific chronic disorders such a generalisation could be dangerous were it to be made the basis for the formulation of a policy for aid, for instance. Certainly a comparison of the two groups of chronically ill infants in this study would belie that generalisation.

Although the undiagnosed coeliac infant may be cross and unhappy, once his diet is established satisfactorily he has then every opportunity of having a normal infancy so long as his mother enforces and he learns to obey the dietary rules. For the fibrocystic infant the problems are not so easily solved. This young child has to have medicines which may taste unpleasant and

physiotherapy which may be uncomfortable. Even so he may have recurrent problems, e.g. bowel prolapse and he is certainly constantly at risk of catching chest infections; these problems expose him to the risk of having the most traumatic experience of all, that of being admitted into hospital away from his mother. The physical symptoms and their treatment then, create a vastly different environment for the infancy of the fibrocystic from that in which the coeliac infant is reared. Objectively then the situation for all chronically ill infants is not the same.

On a more subjective plane the state of the mother should be recalled. Although the mother of the coeliac infant may have difficulties in mastering the child's gluten-free diet and may be as likely as the mother of the C.F. child to request help in the early weeks her needs are fairly readily met. During the infancy of the fibrocystic child his mother is living in the terror of the knowledge that her child has a potentially fatal disease and in the early stages she is likely to be afraid that any mistake that she may make in carrying out his arduous treatment regimen may threaten his life. The nature of the child's symptoms have concerned two of the fundamental activities of infancy, feeding and toilet training and the uncertainty about where the influence of the disease ends and that of the individual child begins makes mothers very ambivalent in their handling of these issues.

At the same time the mother of the C.F. infant is likely also to be concerned with the genetic implications of the child's disease and perhaps preoccupied with some conflict over future family planning.

It is not surprising then that the combined effect of all these stresses should create a situation in which these children are more likely to present infant problems than are their coeliac contemporaries. Clearly then at this stage chronicity of the child's disease is not the most important factor.

Social Adjustment

Although the Vineland Social Adjustment Scale (Doll, 1953) have its limitations the most serious of these from the point of view of this study was the lack of British norms. Even with the use of the control infants for comparison there remained some doubt as to the meaning of the numerical scores and the validity of the scale from which they were derived.

Nevertheless, from the point of view of this study provided a useful basis for the discussion of the development of the chronically ill infants in this study. The most interesting finding to emerge from the comparisons of the groups in these terms did not, in this small sample reach statistical significance but it is a point to be noted for future investigation with larger samples.

The tendency certainly existed for the C.F. infants to lag behind in the development of socialisation. Parents, weighing up the benefits of social contacts against the risks that the child would catchinfections, become physically exhausted or be unable to defend himself in a squabble, in some cases opt for the safe course and this seemed to represent the beginning of a long lonely childhood for these children. It is in matters of this nature that it was felt that parents who had successfully

brought their children through this stage as normally as possible could give confidence and encouragement to those whose children were still young although the difficulties of maintaining an atmosphere of optimism among a group of C.F. parents have already been acknowledged. In cases such as cystic fibrosis then when parents may not come into contact with other parents it may fall to the paediatrician or to our already overworked home visitor to discuss these issues with parents. The main point to be made from the point of view of this Section is worth repeating then, among chronically ill infants the concomitants of the condition for the family are shaped by not only the chronicity of the disease but by several other important considerations e.g. prediagnostic experiences, clinical features treatment, genetic implications and of course, prognosis.

Children and their Illness

The response of children to their knowledge and experience of their own chronic illness shows but one facet of the influence of that disease upon their lives but it may again help to make the point that additional factors besides chronicity are important in shaping the concomitants of chronic disease in childhood.

In the case of coeliac disease the child is likely to be informed from an early age, of his condition and of the treatment required. It is necessary that he should be so informed for his intelligent cooperation is vital to the success of the treatment. As a young infant he may protest violently at being

denied certain foods and as an older child he may certainly be embarrassed by having to have his own food on social occasions. In many cases, the mother's cautionary words may be reinforced by the child's experience of being ill after a dietary indiscretion. With the help of his parents then the coeliac child can learn to overcome his natural resentment of his lot and indeed by the time of the study many of these children were able to consider their situation with stoic calm and resignation.

The position of the C.F. child is rather different. His symptoms are likely to be more troublesome and histreatment more unpleasant. Admissions to hospital may be required and the procedures they involve may be painful or frightening. Yet throughout these experiences the explanation given may be scant. When it is given it may be distressing or at least bewildering for we must acknowledge these are hard facts to make palatable to a child. Nevertheless the most critical difference between these two groups can be related to their differing prognoses. The devastating effect which the knowledge of the prognosis can have on these children has already been acknowledged (Section IV) but even when the child is not explicitly aware of the possible outcome of his condition he may sense it in the responses and attitudes of his parents. Their silence may be as anxiety-provoking as the information they have to impart.

Without labouring the point then, the findings of this study support the contention that chronically ill children are, by virtue of their chronic illness, placed in a situation which is likely to create personal and social problems for them. The results show that some of these problems may cut across the

experience of children with different disorders but they also point out the radical difference which is brought to the child's situation, both directly in his own reactions and indirectly through the reactions of others, when his chronic illness has a fatal prognosis.

The Chronically Ill Child at School (Section V.E.3.)

The point which is to be made from the findings of this part of the study is not the obvious one that the child who is ill and/or who is frequently off school is unlikely to do as well as his capabilities would otherwise allow. Clearly this is true and it is more likely to be applicable to C.F. children than to coeliacs. The matters at issue here are concerned with the less direct concomitants of the child's disease.

A potentially dying child presents a problem to parents in relation to educational values. In relation to other illnesses or handicaps the question of providing some sort of training within the individual's capabilities may be fairly readily solved but the situation of the child who has all his faculties but a limited time in which to develop them may pose something of a dilemma. It might be expected that this would be particularly true of the "future-oriented middle class" (Newson and Newson, 1970) for whom educational values take a prominent position in the ethic of postponement of present pleasure for later benefit. In the face of this situation it is not surprising if parents place less emphasis on school work

for the C.F. child and if in consequence he attaches less importance to scholastic achievement. Although clearly this does not happen in all cases views of this nature were expressed sufficiently frequently for the point to be made that in the context of educational values the prognosis and not only the chronicity of the child's disease is an influential factor.

Attainment Testing

It is acknowledged that the business of comparing intelligence and performance scores is a rather outmoded approach to testing the intellectual attainment of school children within the constraints of the study it seemed the only possible means of obtaining the desired information but the findings are unsatisfactory.

In view of the minor revolution which has taken place in primary school teaching in particular the whole approach of the tests used was strange to some of the children, placing those at more traditional primary schools at something of an advantage. The materials contained within these tests were not universally relevant to all the children as we have already indicated, since variations in curricula between schools introduced an uncontrolled source of variance into the situation. Finally, the testing conditions were variable. Although the secure atmosphere of the child's home was preferable to singling the child out of a class room or testing him in a clinic and was administratively simpler, more informative and more economical than testing his whole class, it also left the investigator rather less control

over the situation. In some cases where parents remained in the room the children were inhibited by their presence, particularly if they reacted in any way to the child's responses. Although the parent who tried to prompt the child was rare the children soon learned to refer to these parents for reinforcement after each response. Cystic fibrosis parents tended to be more interested than those of the control group in the fruits of this enquiry and were more likely to remain in the room during the testing procedure.

Although testing of the children was carefully carried out and concessions were made in the scoring to attempt to combat these sources of error the author is not entirely satisfied with these results. Nevertheless the trend which these results indicate is thought to be a valid one. More factors militate against the C.F. child's fulfilment of his intellectual potential so it is not surprising that this should be the case.

Social and Emotional Development. (Section V.E.5.)

It has already been suggested both in Section IV and in relation to the discussion of the development of infants, in this section, that cystic fibrosis can have an indirect but observable influence on the social development of these children. The reports of mothers in the control group giving comparative accounts of the social development of their coeliac children endorse this view by the contrast which they present. The tendency to timidity and social isolation among C.F. children was seen to stem from two sources, from the children's own natures and from the attitudes of their parents.

The distinction between the two groups was rather less pronounced when the children's school behaviour was compared. Here the tendency was for coeliac children to be described as distractible, restless and impulsive. In some cases such inconsequential behaviour was also associated with anti-social or hostile attitudes. Both boys and girls tended to show these over-reactive behaviour patterns. Although it was thought possible that such behaviour might be an expression of poor dietary control it was evident that the child's knowledge of his illness was also an influential factor.

Knowledge of cystic fibrosis, it will be recalled, tends to evoke quite different behaviour patterns and the tendency there is toward unforthcoming, withdrawn and depressed behaviour.

The numbers of children were rather small for satisfactory statistical analysis and there were some doubts about the weight which could be attached to the numerical scores so that interpretation of these findings has been based more upon the items scored rather than upon the marks thus obtained. On this basis there is a suggestion worthy of further investigation that the clinical features and the prognosis of the child's condition may be important mediators of the influence of that disease on the social development of the affected child.

Social Adjustment

Although the value of the British Social Adjustment Guide was defended in Section II some reservations about its use in this study need to be acknowledged here.

In the first instance there were enormous variations in the standards of behaviour which were considered to be the norm in different schools and the environment against which the school behaviour of the chronically ill child was to be reviewed was thought to be a significant factor in the assessments made. Although local controls were also assessed by means of the Guide, in an effort to combat this source of variance they in themselves seemes to complicate rather than to simplify the situation in some cases.

Another problem was encountered in the teachers' attitudes. If the teacher took a definite stand on the matter of the education of the child in question, then this was likely to be reflected in her scoring of the Guide. For example, in spite of repeated assurances to the contrary, one or two teachers suspected that the study was designed to assess the appropriateness of educating C.F. children in normal schools. These teachers then gave glowing accounts of the children, which were felt to be rather biased. On other occasions, teachers may have been anxious to provide the information which was required and, clearly feeling that a too perfect assessment did not fit the bill, they would endorse items included in Stott's scale, but then would qualify them with copious notes in the margin, making it hard to determine whether or not the item should be scored. Even with the most objective approach possible in such a subjective system of assessment the halo effect was hard to avoid, and there were cases when so many items were endorsed that it was rather hard to believe that the child in question could be so bad. This tendency to high scores was noted too in the control groups of supposedly normal children, as well as among the descriptions of the chronically ill children and

their siblings. It would seem to be a danger in this method of assessment that what is scored is a concept of the child not necessarily his behaviour. The desire for consistency then and the operation of the halo effect may then boost the child's scores on these scales far beyond that which is objectively deserved.

The problem is that these were dangers which were recognised. They were not known for certain to occu and if they did occur, the extent of the bias which they introduced is not known. This is not to invalidate these findings altogether but to note that too much emphasis should not be placed upon the figures quoted, but rather, attention should be given to the trends suggested. The experience of this study would seem to indicate a need for an investigation of the vulnerability of Stott's scales to such extraneous factors.

Emotional Aspects and Older Children (Section V.E.4.)

The adolescence of the normal child is likely to be fraught with emotional difficulties. The deviant youth who has a chronic illness at this time is understandably more likely to be at risk. The sequelae of chronic illness then, may fairly be said to cut across the boundaries of specific disorders at this stage. This thesis however seeks to rederess the balance from a tendency to overgeneralisation, by pointing out particular emotional hazards which exist for C.F. adolescents. From this comparative study, two additional factors in their chronic illness seem to emerge as being particularly threatening to the emotional stability of young people with cystic fibrosis, at this stage.

The retardation of growth and sexual development which characterises cystic fibrosis seems to intensify the unhappiness of young people at this time, increasing their feelings of deviance and their tendency to social isolation. This finding, particularly relevant to C.F. boys, was endorsed by the reports of other researchers e.g. Teicher, 1969; McCollum and Gibson, 1970. On this basis then, it may be suggested that the nature of the clinical features of the chronic disease may indeed be influential, particuarly in the emotional reactions of the afflicted adolescent.

The factor of the prognosis is seen to be even more influential and for the problems it arouses there are no ready solutions. Certainly the C.F. adolescent shares the physical, social and emotional problems of other chronically ill or disabled young people but he has an additional and heavier cross to bear.

In becoming aware of their prognosis it is understandable that these young people may be overwhelmed by feelings of hopelessness and despair. Their parents, weighed down by their own anxieties and fears may be powerless to help them. It is in this environment that the feelings describing the futility of treatment may be expressed by the child and condoned by the parent (McCollum and Gibson, 1970), suicide attempts may be made (Patterson, 1969) and anti-social behaviour patterns exhibited (Teicher, 1969). Although such extremes of reaction were not observed in the sample under study, their precursors of despondency and depression were noted and found to recur at intervals among these young people.

Surely then, these findings indicate a special vulnerability

created by the continuing danger that the child may become ill and die of his chronic illness. No miraculous measures can be suggested which will spare these children or their families from the direct concomitants of this most devastating fact. In some respects those who do not react against its influence are in fact in more danger than those who do. The most expedient course that can be suggested by the findings of this study stems from a need for the early recognition of the special hazards created by the chronic illness which has a potentially fatal prognosis. A willingness to tackle from the first, all the psychological stresses which are thus imposed on the child and his family may at least create a strong position for them from which they may be better able to tackle this most thorny emotional problem which avails them in the child's adolescence.

The Healthy Siblings (Section V.F.)

This study of the behaviour of the healthy siblings of chronically ill children was not definitive but it suggested trends worthy of further investigation. The tendency for these siblings to be attention-seeking was exemplified in the school behaviour of both groups, to a degree which suggested that the element of the situation to which the children were responding was similar in the two cases. However notable differences were identified in the behaviour of the children in their homes, toward their ill sibling.

It is a mark of the normalcy of the coeliac children that they are not cast into the sick role and their condition is

is unlikely to intervene in their relationships with their siblings. The exception to this is clearly in the matter of food where jealousies may be readily aroused by any suggestion of favouritism being shown to either side. In some cases older siblings may be charged to watch over the food intake of the coeliac child outside the home but this increased sense of responsibility for the ill child was not a dominant feature. These findings were in contrast to the findings in the C.F. families where the seriousness of the situation was mediated to the children through their parents' reactions and through the events that befell the sick child. Siblings of C.F. children were more likely then to be disturbed by the illness of their sibling.

Since the study of siblings had not been a detailed one and since the behaviour of the siblings of these C.F. children did not appear to be significantly different from that described by McMichael (1971) among the siblings of children with a variety of physical handicaps. This is not an area in which the special influence of the child's prognosis can be stressed.

In this Section the findings of the two studies of chronically ill children were compared and discussed with respect to the importance of the factor of their differing prognoses in the observed psychological concomitants of each.

This study however, began with a number of broad aims, and in conclusion we need now to consider the extent to which these findings satisfy the goals that were set.

SECTION VII SUMMARY AND CONCLUSIONS

A review of the literature reveals that in the past there have been several studies which have attempted to evaluate the psychological concomitants of chronic disabilities or disfigurements. Few of them however, have considered the sequelae of chronic illnesses which do not present a physical handicap or a visible stigma. Cystic fibrosis is just such a chronic illness. In most cases it does not impose a grave physical handicap nor does it maim the afflicted children, but it is a potentially fatal disease and, as such, might be expected to have serious psychological implications both for them and for their families. In the past there has been little opportunity for assessment of the nature or extent of these implications, since the children tended to die in their early years. Now modern medicine has improved the life expectancy of fibrocystic children to the point where such an investigation not only becomes possible, but becomes necessary, in order that psychological problems occasioned by the child's condition may be identified and, as far as possible, alleviated.

Some research workers had already tackled this problem but their published reports showed some important shortcomings of which note was taken in the design of this study.

50 families with 58 fibrocystic children took part in this investigation. The families were all National Health Service patients so that the problem of the anxiety aroused by the financial stress of paying for long term medical care, which was a dominant factor in these previous studies, was minimised.

Information was gathered by interviews and psychometric tests carried out in the families' own homes. It was hoped, by this means to provide a fairer assessment of their emotional state than had been given by previous studies which had gathered data exclusively within the anxiety-provoking setting of a hospital clinic. Mothers, fathers and the children themselves contributed to the study which attempted to provide a more global picture of the situation of the family affected by cystic fibrosis, than previous studies had done. School teachers provided additional information about the behaviour of school children in the sample outwith their own homes.

The study had two further methodological advantages over previous studies in that a larger sample of C.F. children was available and that the findings of the study were evaluated with reference to a control group. The control group was selected from children suffering from coeliac disease. This disease is similar to cystic fibrosis in being chronic and incurable, yet neither disabling nor disfiguring and, as in cystic fibrosis, responsibility for its day-to-day treatment falls largely on the children's parents. However, coeliac disease, unlike cystic fibrosis, does not have a grim prognosis. A sample of coeliac children was selected then, to correspond to the sample of C.F. children as closely as possible in terms of:

(a) the child's sex

(b) the child's age

(c) the size of the child's family

(d) the socio-economic status of his family.

Each family in each of the two groups was visited in its home on several occasions. At the first visit an interview was conducted, usually with the child's mother, to provide some biographical data about the family. The child's medical history was recorded and the family's experience of his medical treatment and supervision was explored, with particular attention being given to unfulfilled needs for help. Practical, social and economic problems raised by the child's illness were investigated and the influence of the child's disease on the attitudes of others to him was probed. The child's personal development from birth to the time of the study was traced. On the second visit the more personal aspects of the mother's response to the child's illness were considered and her account of the concomitants of the disease for her family was recorded. Corresponding information was obtained from the child's father on a separate occasion. Each parent was invited to complete a personality questionnaire, Cattell's 16 P.F., and an anxiety inventory, Taylor's Manifest Anxiety Scale.

Information about the development of the youngest children re those under 5:0 years was gathered from their mothers with the aid of the Vineland Social Maturity Scale. Older children took a more active part in the study. The intelligence and educational attainment of the school children in each group was assessed by means of standardised tests i.e. W.I.S.C., Vernon's Graded Arithmetic-Mathematics Test and Schonell's GradedWord Reading and Graded Word Spelling tests. The older children were interviewed, particularly with a view to discovering their knowledge of and reactions to their illness. They were invited to complete the

Children's Form of the Manifest Anxiety Scale.

The healthy siblings in these families were not studied directly although parents' views of their general behaviour and in particular of their reaction to their ill sibling were recorded.

Where either the ill children or their siblings were still at school, an additional source of information was available. Teachers' assessments of these children's school behaviour were gathered, with the aid of Stott's Bristol Social Adjustment Guide for the 'Child in School'.

An attempt was made to evaluate the findings of the study at two levels : the theoretical and the practical. At the theoretical level the importance of the factors of the visibility of the disease and of its prognosis were considered.

It had been anticipated that the lack of visible physical stigma in most cases of these two diseases would create the possibility of rather different psychological concomitants from those of the disfiguring conditions previously studied. Certainly the absence of overt disfigurement allowed the child or his family the option of 'passing' (Goffman, 1968), if they did not want others to know of the child's condition. It also allowed more room for doubt of the diagnosis in some families. However, for some of the children, physical signs were, or were felt to be, evident e.g. in thinness or abdominal distension, while in other cases, either the symptoms of the disease or their treatment, or both, proved to be socially inconvenient in a way which was just as stigmatising as the effect of the truly visible physical abnormalities. To some extent then, these children were still differentiated from their peers and hence more similarities than differences were noted,

on this basis, between the findings of this and previous studies. In this respect then, we may support the tenet that the shared factor of the chronicity of the diseases investigated is a more important influence in shaping their psychological sequelae than is the distinguishing feature of their differing degrees of visibility.

The influence of the prognosis was a rather different matter, which was assessed by comparison of the findings from the C.F. group with those from the control group. The life-threatening nature of cystic fibrosis was found to be a powerful factor, influencing both the nature and the degree of psychological concomitants experienced by families in the two groups.

From the first, the child's prognosis was shown to have considerable impact upon his parents' response to the diagnosis. Thereafter, communications within the family, the mental health of the parents and the personal development of the ill child were found to be particularly vulnerable to the influence of this factor. Thus, it was argued that over and above the common experience of families with chronically ill children, the factor of the child's prognosis exerts a crucial influence. Families of children whose chronic disease is potentially fatal need special care and help, beyond that which is required to support families of children with other chronic conditions.

At the level of practicalities many points of interest arose during the course of the study but only the major issues need be reiterated here.

It was observed that a child's chronic illness begins to influence the life of his family, often, even before it is diagnosed

and it was suggested that more attention be given to the events of this pre-diagnostic period. In this study evidence was found of a need for more information, particularly about cystic fibrosis, to be disseminated to doctors in the community, to hasten the diagnosis of this disease. Delays and difficulties during the pre-diagnostic period were found to be influential, not only in parents' subsequent relationships with their family doctor but also in their emotional state at the time of the child's eventual diagnosis. Greater consideration for parents' prediagnostic experiences in the management of their child's hospital admission and diagnosis was called for.

A diagnosis of cystic fibrosis is inevitably a distressing event for parents' and, in order to avoid exacerbating parents' difficulties at this time, it was suggested that particular care should be taken in the communication of such a diagnosis. Attention was drawn to the importance of the practical aspects of the management of this communication; specifically, the need for the diagnosis to be given to both parents together was particularly stressed. Problems in the explanation of medical matters to lay people were noted and in particular, the difficulty of striking a balance between hopeless pessimism and unrealistic optimism in the explanation of the child's prospects, was acknowledged. Parents expressed a preference for a simple, honest explanation which prepared them for the worst but left some room for hope. Nevertheless, parents were naturally shocked by this information and a need for repeated opportunities to discuss the diagnosis with the child's doctor was evident. Opportunities for such encounters, which should be initiated by the doctor, were found

to be particularly valuable when they encompassed consideration of parents' emotional responses as well as the hard facts of their child's diagnosis. Parents' early involvement in the child's treatment, even while the child was still in hospital, was seen to be useful in helping to combat their feelings of helplessness and lack of confidence at this time.

Although coeliac mothers were found to have some needs in common with C.F. mothers at this time, e.g. for repeated explanations or for opportunities to participate in the child's treatment under the hospital's supervision, their situation in emotional terms, was markedly different. The findings from the control study then, highlighted the special problems in the management of a diagnosis of a potentially fatal disease of childhood.

Parents' understanding of their children's disease was found to be generally poor, in view of their high degree of involvement in its treatment. The ignorance of the general public in matters of human physiology and the difficulty experienced by some medical men in communicating in lay terms were seen to contribute to this problem. It was suggested that the preparation of written information for parents, to endorse the doctor's verbal communication of the child's diagnosis, might alleviate this difficulty.

A special problem here was evident in the genetic aspects of cystic fibrosis, of which parental comprehension was particularly poor. It was seen to be important that parents should be better informed in this matter and that genetic counselling and family planning services should be more readily available to them.

Although on the whole, parents' attitudes were favourable

towards the system of care provided for their child some omissions in this system became apparent in the course of this study. The burden of responsibility for the child's daily treatment weighed heavily on parents and some additional source of assistance was seen to be required, particularly by mothers of fibrocystic children. Needs for information, for advice with problems in relation to the child and for a source of emotional support for parents were identified and a home-visiting service was introduced in an experimental way, in an attempt to meet the needs of these C.F. families.

Cystic fibrosis did not create financial difficulties for many of the families in this sample although the social and interpersonal aspects of family functioning did seem to be influenced by the disease. Some evidence of social isolation of these families was recorded but it was less severe than the isolation described in other studies. The alarming prognosis of cystic fibrosis was found to have an inhibiting effect on family communications, in relation to the child, his illness and his future, and it had repercussions in the attitudes of others to the child. The impact of cystic fibrosis upon family functioning was thus seen to be different from that of coeliac disease.

Cystic fibrosis had a particularly profound effect on the lives of parents of affected children. It was found to intervene in the parent-child relationship, making parents more uncertain in their handling of the ill child. This uncertainty was often resolved in the direction of lowered expectations being held of that child, in a way which was not always commensurate with the severity of his condition. In some families cystic fibrosis

also intervened in the parents' marital relationship. Here, its disruptive influence could be seen in parents' level of communication with each other, in their sexual relationship and in their shared activities, both within the family and outside. Happily, many relationships were found to have been strengthened, rather than broken, by the stresses imposed by cystic fibrosis. Finally, cystic fibrosis was shown to present personal difficulties for parents. The most striking instance of this was seen in its deleterious effect on the mental health of the children's mothers. The influence of cystic fibrosis upon the parents of affected children was thus found to be more severe than that of coeliac disease, a factor which was largely attributable to the uncertainty of its outcome.

In spite of all the problems, many of these mothers coped admirably. Nevertheless, it was clear that any measure which could lighten their burden should be considered. The role of parents' groups was discussed in this context but it was concluded from the experiences of this sample that parents could be more effectively supported and helped by their relationships with their doctors and, perhaps, with the home-visitor, than by uncontrolled contacts with other parents in the same situation as themselves. The situation was found to be rather different in the control group, since the exchange of views between parents of coeliac children is much less likely to be threatening and indeed, is more likely to be practically useful, to these parents.

Cystic fibrosis was acknowledged to present some real problems during the children's development. However, a risk was noted that these difficulties might be compounded by other factors,

less directly related to the child's physical limitations, and such dangers were first identified during the child's infancy. At this time, an exacerbation of the normal infant difficulties could often be seen to be associated with parents' reactions to the child's diagnosis. For the older child, the pattern developed and the risks of social isolation and emotional withdrawal became greater, particularly among boys in the sample. Although the children showed a normal distribution of intellectual capacity, they were frequently found to show evidence of under-achievement in school subjects. With adolescence, the social and adolescent problems became complicated by sexual ones, in particular of the delayed sexual development of fibrocystic young people; at the same time, scholastic problems gave place to vocational ones. The findings of the study suggested that the uncertainty of the future of these children had played a dominant and often a spoiling role, throughout their development. Since the difficulties at each stage seemed to come as a logical progression from earlier problems, it was suggested that the issues concerning the child's personal development needed to be brought to the forefront of awareness from the first. It was suggested that the services available to help these families in the medical management of their children's illness, might expand the scope of their concern to include other aspects of the child's development. It was hoped that the home visiting service might help in this matter by affording parents' a better insight into their child's normal needs as well as into the concessions required by his illness. Once more, the influece of coeliac disease upon the development

of the children in the control group was found to be considerably less disturbing.

The situation of healthy children in these families was not explored in detail. Nevertheless, it was apparent that if the ill child was capturing more than his share of parental time and attention his siblings were likely to show some adverse reactions. Attention-seeking behaviour and feelings of jealousy were most frequently described among younger siblings. Older children, who were often exhorted by the parents to look after their ill sibling, were less likely to show their resentment directly in the home but disturbances in their school behaviour were found to be fairly common. At school the children were often said to be over-reactive. Further study in this area was called for, to elaborate upon the patterns of sibling behaviour hinted at by these findings.

In conclusion then, we should acknowledge that this study began with several weaknesses, not all of which were able to be overcome. Nevertheless, it is hoped that its conduct and its outcome may have gone some of the way to presenting a study of the psychological concomitants of chronic illness in childhood which is theoretically interesting, methodologically sound and practically valuable.

SECTION VIII REFERENCES

ABRAMS, R.C. (1970) The Impact of Cerebral Palsy on the Patient and his Family. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas

ADLER, A. (1927) The Practice and Theory of Individual Psychology New York : Harcourt Brace.

ALLEN, F.H. and PEARSON, G.H.J. (1928) The Emotional Problems of the Physically Handicapped. Brit. J. Med.Psych.,8, 212-235.

ALTMEYER, L. (1969) Observations of a 28-year old man with cystic fibrosis. Minn.Med., 1397-1399.

APLEY, J. and MACKEITH, R. (1962) The Child and his Symptoms : A Psychosomatic Approach. Oxford : Blackwell Scientific Publications.

BAGLEY, C. (1971) The Social Psychology of the Child with Epilepsy London : Routledge and Kegan Paul. International Library of Sociology.

BARKER, R. (1960) Introduction to WRIGHT, B.A. Physical Disability - A Psychological Approach. New York : Harper and Row.

BARKER, R. and WRIGHT, B.A. (1952) The Social Psychology of Adjustment to Disability. In GARRETT, J.F. (Ed.) Psychological Aspects of Physical Disability. 2nd. edn. Washington D.C. : U.S. Govt. Printing Office.

BARKER, R.G., WRIGHT, B.A., MEYERSON, L. and GONICK, M.R. (1953) Adjustment to Physical Handicaps and Illness - A Survey of the Social Psychology of Physique and Disability. Revised edn. New York : Soc.Sci.Res.Council, Bulletin 55.

BARRON, J. (1955) Physical Handicap and Personality : Study of the Seen Versus Unseen Disabilities. Chicago : Archives of Physical Medicine and Rehabilitation, 36, 639-43.

BATTEN, J. Cystic Fibrosis and the Teenager. Bromley, Kent : The Cystic Fibrosis Research Trust. Ref. CFL/2.

BELMONTE, M.M. (1969) Aspects Psychiques et Emotifs Dans La Fibrose Kystique du Pancreas (Mucoviscidose) Union Med. Canada,98,1943-1945.

BERGMANN, T. (1965) with FREUD, A. Children in the Hospital New York : International Universities Press Inc.

BLUMENTHAL, M.D. (1969) Experiences of Parents of Retardates and Children with C.F. Chicago : Arch. Gen.Psychiat., 21, 16-171.

BRUNT, P.W., SIRCUS, W., MACLEAN, N. (1969) Neoplasia and Coeliac Disease. The Lancet, 1, 180-184.

BRUSILOW, S.W. (1970) Cystic Fibrosis in Adults Annual Rev.Med., 21, 99-104.

BURGESS, L. (1969) Morale Boosting in Cystic Fibrosis. Amer.J.Nursing, 69, 322-323.

BURTON, L. (1973) Caring for Children with Cystic Fibrosis. The Practitioner, 210, 247-254.

CAPLAN, G. (1961) An Approach to Community Mental Health New York : Grune and Stratton.

CARTER, C.O. The Genetics of Cystic Fibrosis. Bromley, Kent : The Cystic Fibrosis Research Trust. Ref. CFL/1.

CASTANEDA, A., McCANDLESS, B.R., PALERMO, D.S. (1956) Children's Form of the Manifest Anxiety Scale. Child Development,27,317-326.

CATTELL, R.B., EBER, H.W. and TATSUOKA, M.M. (1970) Handbook for the 16 PF Questionnaire. Champaign, Illinois : Institute for Personality and Ability Testing.

CHAPMAN, D. (1970) One Little Boy with Two Big Problems. Canad. Nursing, 66, 36-38.

CHAZAN, M (1968) Inconsequential Behaviour in School Children. Brit. J.Educ.Psych., 38, 1, 5-7.

CHAZAN, M. (1970) Maladjusted Children. In MITTLER, P. (Ed) The Psychological Assessment of Mental and Physical Handicaps. London : Methven.

CHESLER, M. (1965) Ethnocentrism and Attitudes Toward the Physically Disabled. J. Personality and Soc. Psych., 2, 877-882.

COCHRAN, W.G. (1954) Some Methods for Strengthening the Common Chi-Square Tests. Biometrics, 10, 417-451.

CORMACK, P. (1967) The Relation between Body Cognition and Attitudes Expressed Toward the Visibly Disabled Rehab.Couns.Bull., 11, 106-109.

COWEN, E.L., UNDERBERG, R.P., VERRILLO, R.T. and BENHAM, F.G. (1961) Adjustment to Visual Disability in Adolescence. New York : American Foundation for the Blind.

CRUIKSHANK, W.M. (1951) The Relation of Physical Disability to Fear and Guilt Feelings. Child Development, 22, 291-298.

DAVIE, R. (1968) The Behaviour and Adjustment of Seven Year-Old Children. Brit.J.Educ.Psych., 38, 1, 1-2.

DAVIES, C.M. (1965) Development of the Probability Concept in Children. Child Development, 36, 779-788.

DEBUSKEY, M. (1970) The Chronically Ill Child and His Family. Springfield, Illinois : Charles C. Thomas.

DIBBEN, C. Cystic Fibrosis and the Medical Social Worker. Bromley, Kent : The Cystic Fibrosis Research Trust. Ref.CFL/4.

DOLL, E.A. (1953) The Measurement of Social Competence Minneapolis Educ. Test Bureau.

DOMBRO, R.H. and HAAS, B.S. (1970) The Chronically Ill Child and His Family in the Hospital. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and His Family. Springfield, Illinois: Charles C. Thomas.

DOUGLAS, J.W.B. (1964) The Home and The School. London : Macgibbon and Kee

DOUGLAS, J.W.B. and BLOMFIELD, J.M. (1958) Children Under Five. London : George Allen and Unwin.

DOUGLAS, J.W.B. and ROSS, J.M. (1968) Adjustment and Educational Progress. Brit.J.Educ.Psych., 38, 1, 2-4.

DOW, T. (1965) Social Class and Reaction to Physical Disability. Psych.Reports, 17, 39-62.

DOWNIE, N.M. and HEATH, R.W. (1965) Basic Statistical Methods. London : Harper and Row Ltd.

ELLIOTT, K. (1970) The Family and Its Future. A Ciba Foundation Blueprint. London : J. & A. Churchill.

EMERY, A.E.H., WATT, M.S. and CLACK, E. (1973) Social Effects of Genetic Counselling. Brit. Med.J., 1, 724-726.

ENDLER, N.S., HUNT, J.M. and ROSENSTEIN, A.J. (1962) An S-R Inventory of Anxiousness. Psych.Monographs, 76, No.17, Whole No.536.

EYSENCK, H.J. and RACHMAN, S. (1973) The Future of Clinical Psychology. Bull.Brit.Psych.Soc., 26, 113-116.

FESTINGER, L. (1962) Cognitive Dissonance. Sci.Amer. 207, 93-102.

FESTINGER, L. (1962) A Theory of Cognitive Dissonance. Stanford: Stanford University Press.

FORUCCI, R.A., and STARK, E.W. (1972) Hearing Loss, Speech-Language and Cystic Fibrosis. Arch.Otolaryngol., 96, 361-364.

FOX. R. and PARSONS, T. (1952) Illness, Therapy and the Modern Urban Family. J.Soc.Issues, 8, 31-44.

FRIEDMAN, S.B., CHODOFF, P. and MASON, J.W. (1963) Behavioural Observations on Parents Anticipating theDeath of a Child. Pediatrics, 32, 610-625.

GARDNER, R.A. (1968) Psychogenic Problems of Brain-Injured Children and their Parents. J.Amer.Acad.Child Psychiat., 7,471.

GARDNER, R.A. (1969) Guilt Reactions of Parents of Children with Severe Physical Disease. Amer.J.Psychiat., 126, 636-644.

GARRETT, J.F. (1952) Psychological Aspects of Physical Disability. 2nd. edn. Washington.D.C. : U.S. Govt. Printing Office.

GATES, M.F. (1946) A Comparative Study of Some Problems of Social and Emotional Adjustment of Crippled and Non-Crippled Boys and Girls. J. Genetic Psych., 68, 219-244.

GLASER, H.H., LYNN, D.B. and HARRISON, G.S. (1961) Studies of Comprehensive Medical Care for Handicapped Children. II Patterns of Anxiety in Mothers of Children with Rheumatic Fever. Amer.H. of Diseases of Children 102, 3, 344-354.

GLASER, H.H., HARRISON, G.S., and LYNN, D.B. (1964) Emotional Implications of Congenital Heart Disease in Children. Pediatrics, 33, 367-379.

GOFFMAN, E. (1968) Stigma : Notes on the Management of Spoiled Identity. Harmondsworth : Pelican Books.

HALL, C.S. and LINDZEY, G. (1966) Theories of Personality. 15th print. New York : Wiley.

HALLER, J.A., Jnr. (1970) A Healthy Attitude to Chronic Illness. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

HAMBURG, D.A. and ADAMS, J.E. (1967) A Perspective on Coping Behaviour. Arch.Gen.Psychiat., 17, 277-284.

HARRIS, D.B. (1963) Children's Drawings as Measures of Intellectual Maturity : A Revision and Extension of the Goodenough Draw-A-Man Test. New York : Harcourt, Brace and World Inc.

HARROWER, M. (1955) Medical and Psychological Teamwork in the Care of the Chronically Ill. Springfield, Illinois : Charles C. Thomas.

HAWKES, G.R. and PEASE, D. (1966) Behaviour and Development from Five to Twelve. New York : Harper.

HEWETT, S. (1970) with NEWSON, J. and NEWSON, E. The Family and the Handicapped Child. London : George Allen and Unwin Ltd.

HOFFMAN, L.W. and LIPPITT, R. (1960) The Measurement of Family Life Variables. In MUSSEN, P.H. (Ed.) Handbook of Research Methods in Child Development. New York : Wiley.

HOPKINS, E.W. (1970) The Chronically Ill Child and the Community In DEBUSKEY, M. (Ed) The Chronically Ill Child and His Family. Springfield, Illinois : Charles C. Thomas.

HOMANS, G. (1960) The Human Group. New York : Harcourt Brace.

ILLINGWORTH, R.S. (1972) The Normal Child : Some Problems of the Early Years and their Treatment. 5th edn. Edinburgh : Churchill Livingstone.

JACO, E.G. (1958) Patients, Physicians and Illness. New York : The Free Press.

JOLLY, H. (1968) Diseases of Children. 2nd edn. Edinburgh : Blackwell Scientific Publications.

KAHN, R.L. and CANNELL, C.F. (1957) The Dunamics of Interviewing. New York : Wiley.

KAMMERER, R.C. (1940) An Exploratory Psychological Study of Crippled Children. Psych. Record, 4, 47-100.

KERSHAW, J.D. (1966) Handicapped Children. 2nd edn. London : Heinemann Medical Books.

KULCZYCKI, L.L. (1970) Adequate Home Care for Patients with Cystic Fibrosis. Clin.Proc.Child.Hosp.D.C., 26, 97-103.

KULCZYCKI, L.L., ROBINSON, M.E. and BERG, C.M. (1970). Somatic and Psychosocial Factors Relative to Management of Patients with Cystic Fibrosis. Clin.Proc. Child Hosp. D.C., 25, 320-324.

LANGE, P. (1959) Frustration Reactions of Physically Handicapped Children. Exceptional Children, 25, 355-357.

LAWLER, R.H., NAKIELNY, W. and WRIGHT, N.A. (1966). Psychological Implications of Cystic Fibrosis. Canad.Med.Assoc.J., 94, 1043-1046.

LEONARD, C.O., CHASE, G.A. and CHILDS, B. (1972) Genetic Counselling : A Consumer's View. New Eng.J.of Med., 287, 9, 433-439.

LEVIN, S. (1970) Nephrotic Syndrome in Childhood. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

LEVITT, E.E. (1968) The Psychology of Anxiety. London : Staples Press, Granada Publishing Ltd.

LINDER, R. (1970) Mothers of Disabled Children - the Value of Weekly Group Meetings. Develop.Med.Child.Newrol., 12, 202-206.

LUKENS, J.N. and MILES, M.R. (1970) Childhood Leukaemia - Meeting the Needs of Patient and Family. Missouri. Med., 67, 236-241.

LUKOFF, I. and WHITEMAN, M. (1964) Attitudes Toward Blindness New York : Amer.Fed.Catholic Workers for the Blind.

LUNZER, E.A. (1966) The Manchester Scales of Social Adaptation. Slough : N.F.E.R.

McCOLLUM, A.T. and GIBSON, L.E. (1970) Family Adaptation to the Child with Cystic Fibrosis. J. Paediatrics, 77, 571-578.

McCRAE, W.M. (1969) Inheritance of Coeliac Disease. J. Med.Genetics, 6, 129-131.

McCRAE, W.M. (1973) Personal Communication.

McCRAE, W.M., CULL., A.M., BURTON, L. and DODGE, J. (1973) Cystic Fibrosis: Parents' Response to the Genetic Basis of the Disease. The Lancet, 2, 141-143.

McCREARY, J.B. and BENDIG, A.W. (1954) Comparison of Two Forms of the Manifest Anxiety Scale. J.Consult. Psych., 18, 206.

McDANIEL, J.W. (1969) Physical Disability and Human Behaviour. New York : Pergamon Press Inc.

McMICHAEL, J.K. (1971) Handicap : A Study of Physically Handicapped Children and their Families. London : Staples Press, Granada Publishing Ltd.

MAHL, G.F. (1971) Psychological Conflict and Defense. New York : Harcourt Brace Jovanovich Inc.

MARTORANA, A.A. (1954) A Comparison of the Personal, Emotional and Family Life Adjustment of Crippled and Normal Children. Unpublished Ph.D. Thesis, Univ. of Minnesota.

MENDELSOHN, R.S. (1970) Broadening the Horizons of Pediatric Practice. Problems of Management of Chronic Diseases as exemplified by Cystic Fibrosis. Clin.Pediatrics (phila.), 9, 638-641.

MENG, H. (1938) Schweizer Archiv. für Neurologie und Psychiatrie, 40, 328-344. Translation in BARKER, R. et al (1953) Adjustment to Physical Handicaps and Illness. New York : Soc.Sci. Res.Council. Bull. 55.

MEYERSON, L. (1948) Physical Disability as a Social Psychological Problem. J.Soc.Issues, 4, 2-4.

MITCHELL, S. (1965) A Study of the Mental Health of School Children in an English County. Unpublished Ph.D. Thesis, Univ. of London.

MITCHELL, S. and SHEPHERD, M. (1966) A Comparative Study of Children's Behaviour at Home and at School. Brit. J.Educ.Psych., 36, 248-254.

MITTLER, P.J. (1970) Handicapped Children : Some Common Factors. In MITTLER, P. (Ed.) The Psychological Assessment of Mental and Physical Handicaps. London : Methven.

MITTLER, P. (1970) The Psychological Assessment of Mental and Physical Handicaps. London : Methven.

MOOS, J. and SOLOMON, G. (1965) Personality Correlates to the Degree of Functional Incapacity of Patients with Physical Disease. J.Chron.Dis., 18, 1019-1038.

MUSSEN, P.H. (1960) Handbook of Research Methods in Child Development. New York : Wiley.

MUSSEN, P.H. and BARKER, R.G. (1944) Attitudes Towards Cripples J. Abnorm. and Soc.Psych., 39, 351-355.

NAGY, M. (1948) The Child's Theories Concerning Death. J.Genetic Psych., 73, 3-27.

NATTERSON, J.M. and KNUDSON, A.G. (1960) Observations Concerning Fear of Death in Fatally Ill Children and their Mothers. Psychosom.Med., 22, 6, 456-466.

NEILL, C.A. (1970) The Cardiac Child and his Family. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

NEWSON, J. and E. (1963) Infant Care in an Urban Community. London : George Allen and Unwin Ltd.

NEWSON, J. and E. (1968) Four Years Old in an Urban Community. London : George Allen and Unwin.

NEWSON, J. and E. (1970) Changes in Concepts of Parenthood. In ELLIOTT, K. (Ed.) The Family and its Future. A Ciba Foundation Blueprint. London : J. & A. Churchill.

OAKESHOTT, E. (1973) The Child Under Stress. London : Priory Press Ltd.

OPPENHEIM, A.N. (1966) Questionnaire Design and Attitude Measurement. London : Heinemann Educational Books Ltd.

PARSONS, T. (1958) Definitions of Health and Illness in the Light of American Values and Social Structure. In JACO, E.G. (Ed.) Patients, Physicians and Illness. New York : The Free Press.

PATTERSON, P.R. (1969) Social Stigmata of Cystic Fibrosis Minnesota Med., 52, 1556-1558.

PATTERSON, P.R., DENNING, C.R. and KUTSCHER, A.H. (1973) Psychosocial Aspects of Cystic Fibrosis. Columbia : Univ.Press (in press).

PINKERTON, P. (1969) Managing the Psychological Aspects of Cystic Fibrosis. Arizona Med., 26, 348-351.

PINKERTON, P. (1970) Parental Acceptance of the Handicapped Child. Develop.Med.Child Neurol., 12, 207-212.

PLESS, I.B. and DOUGLAS, J.W.B. (1971) Chronic Illness in Childhood : Part 1. Epidemiological and Clinical Characteristics. Pediatrics, 47, 2, 405-414.

PLESS, I.B. and ROGHMANN, K.J. (1971) Chronic Illness and its Consequences - Observations Based on Three Epidemiologic Surveys. J. Pediatrics, 79, 3, 351-359.

POGUE, R.E. and WARWICK, W.J.(1969) Cystic Fibrosis : A New Challenge to Internal Medicine. Minessota Med., 52, 1551-1555.

PRINGLE, M.L.K. (1964) The Emotional and Social Adjustment of Blind Children. London : N.F.E.R. in England and Wales. Occasional Publication No. 10.

PRINGLE, M.L.K. (1964) The Emotional and Social Adjustment of Physically Handicapped Children. London : N.F.E.R. in England and Wales. Occasional Publication No. 11.

PRUGH, D.G. (1951) A Preliminary Report on the Role of Emotional Factors in Idiopathic Celiac Disease. Psychosom.Med., 13, 4, 220-241.

REYNALL, J. (1970) Children with Physical Handicaps. In MITTLER, P. (Ed.) The Psychological Assessment of Mental and Physical Handicaps. London : Methven.

ROBERTSON, J. (1958) Young Children in Hospital. London : Tavistock Publications.

ROBSON, C. (1973) Experiment, Design and Statistics in Psychology. London : Penguin Education.

RODRIGUEZ, A. (1970) Variation on a Theme. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

ROGHMANN, K.J. and HAGGERTY, R.J. (1970) Rochester Child Health Surveys : 1. Objectives, Organisation and Methods. Medical Care, 8, 47. Reported by PLESS, I.B. and DOUGLAS, J.W.B. (1971)op.cit.

ROSENBAUM, S.Z. (1943) Infantile Paralysis as the Source of Emotional Problems in Children. Welfare Bull., 34, 11-13.

ROSENSTEIN, B.J. (1970) Cystic Fibrosis of the Pancreas - Impact on Family Functioning. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

RUESCH, J. (1946) Chronic Disease and Psychological Invalidism. New York : Amer. Soc.Res. Psychosom. Probs.

RUNCIMAN, W.G. (1963) Social Science and Political Theory. Cambridge University Press.

RUTTER, M. (1967) A Children's Behaviour Questionnaire for Completion by Teachers : Preliminary Findings. J. Child Psych., and Psychiat., 8, 1 - 11.

RUTTER, M. (1970) Psychological Development - Predictions from Infancy. J. Child Psych. and Psychiat., 11, 49-62.

RUTTER, M. and GRAHAM, P. (1966) Psychiatric Disorders in 10 - and 11 - Year Old Children. Proc.Roy.Soc.Med., 59, 382-387.

RUTTER, M., TIZARD, J. and WHITMORE, K. (1970) Education, Health and Behaviour. London : Longmans, Green and Co.

SARASON, I.G. (1960) Empirical Findings and Theoretical Problems in the Use of Anxiety Scales. Psych.Bull., 57, 403-415.

SAVILLE, P. (1971) A British Supplement to the Manual of the Wechsler Intelligence Scale for Children. Windsor, Berks. : N.F.E.R. Publishing Co. Ltd.

SAVILLE, P. (1972) The British Standardisation of the 16 P.F. Windsor, Berks. : N.F.E.R. Publishing Co. Ltd.

SCHACHTER, S. (1961) The Psychology of Affiliation. London : Tavistock Publications.

SCHILDER, P. (1950) The Image and Appearance of the Human Body. New York : Wiley.

SCHONELL, F.J. (1942) Backwardness in the Basic Subjects. Edinburgh : Oliver and Boyd.

SCHONTZ, F.C., FINK, S.L. and HALLENBECK, C.E. (1960) Chronic Physical Illness as Threat. Arch.Phys.Med. & Rehab., 41, 143-148.

SEARS, R.R., MACOOBY, E.E. and LEVIN, H. (1957) Patterns of Child Rearing. Evanston : Row, Peterson.

SEARS, R.R., WHITING, J.W.M., NOWLIS, V. and SEARS, P.S. (1953) Some Child Rearing Antecedents of Aggression and Dependency in Young Children. Genet.Psych.Monogr., 47, 135-234.

SIDNEY, E. and BROWN, M. (1961) The Skills of Interviewing. London: Tavistock Publications Ltd.

SIBINGA, M.S. and FRIEDMAN, C.J. (1971) Complexities of parental understanding of phenylketonuria. Pediatrics, 48, 216-224.

SIEGEL, S. (1956) Non-Parametric Statistics for the Behavioural Sciences. Tokyo : McGraw Hill International Student Edition.

SIGLER, A.T. (1970) The Leukaemic Child and His Family : An Emotional Challenge. In DEBUSKEY, M. (Ed.) The Chronically Ill Child and his Family. Springfield, Illinois : Charles C. Thomas.

SILLER, J. (1963) Reactions to Physical Disability. Rehab.Couns. Bull., 7, 12-16.

SILLER, J. and CHIPMAN, A. (1964) Factorial Structure and Correlates of the Attitudes Toward Disabled Persons Scale Educ. Psych.Meast., 24, 831-840.

SIMPSON, E.E. School Problems of Children with Cystic Fibrosis Bromley, Kent : The Cystic Fibrosis Research Trust. Ref.CFL/5.

SMITH, J. and FINK, S. (1963) The Relationship Between Physical Improvement and Psychological Factors in Chronically Ill Patients. J.Clin.Psych., 19, 289-292.

SPEER, F. (1970) Allergic Children and Parental Attitudes. Clin. Ped.(phila.), 9, 642-647.

SPOCK, A. and STEDMAN, D.J. (1966) Psychologic Characteristics of Children with Cystic Fibrosis. N. Carolina Med.J., 27, 426-428.

SPOCK, B. and LERRIGO, M. (1965) Caring for your Disabled Child. New York : Macmillan.

STACEY, M., DEARDEN, R., PILL, R. and ROBINSON, D. (1970) Hospitals : Children and their Families. London : Routledge and Kegan Paul.

STADNYCK, S. (1970) A Camp for Children with Cystic Fibrosis. Amer.J.Nrs., 1691-1693.

STOLZ, L.M., (1967) Influences on Parent Behaviour. London: Tavistock Publications Ltd.

STOTT, D.H. (1971) The Social Adjustment of Children. 4th.edn. London : Univ. of London Press.

SUTTON-SMITH, B. and ROSENBERG, B.G. (1970) The Sibling. New York : Holt, Rinehart and Winston Inc.

RAUSSIG, L.M., LOBECK, C.C., KATTWINKEL, J. and di SANT'AGNESE, P.A., (1972) Fertility in Males with Cystic Fibrosis. The Lancet, 1,850.

TAYLOR, J. (1953) A Personality Scale of Manifest Anxiety. J. Abnorm. and Soc.Psych., 48, 285-290.

TEICHER, J.D. (1969) Psychological Aspects of Cystic Fibrosis in Children and Adolescents. Calif.Med., 110, 371-4

TEICHER, J.D. (1969) Psychological Aspects of Cystic Fibrosis in Children and Adolescents. Calif.Med., 111, 408-409.

TERMAN, L.M. and MERRILL, M.A. (1961) Stanford-Binet Intelligence Scale. Manual for the Third Revision Form L-M. London : Harrap.

TIZARD, J. (1968) Questionnaire Measures of Adjustment. Brit.J. Educ.Psych., 38, 1, 9-13.

TIZARD, J. (1971) Preface to McMICHAEL, J.K. Handicap. London : Staples Press.

TIZARD, J. and GRAD, J.C. (1961) The Mentally Handicapped and their Families. London : Oxford University Press.

TROPAUER, A., FRANZ, M.N. and DILGARD, V.W. (1970). Psychological Aspects of the Care of Children with Cystic Fibrosis. Amer.J.Dis. Child., 119, 424-432.

TURK, J. (1964) The Impact of Cystic Fibrosis on Family Functioning. Pediatrics, 34, 67-71.

U.S. CHILDREN'S BUREAU (1952) Emotional Problems Associated with Handicapping Conditions in Children. Washington D.C. : U.S. Govt. Printing Office.

VALENTINE, C.W. (1970) The Normal Child. 7th Reprint. Harmondsworth: Penguin Books Ltd.

VERNON, P.E. (1960) Intelligence and Attainment Tests. London : Univ. of London Press.

VERNON, P.E. (1969) Personality Assessment: A Critical Survey : London: Methven, Soc.Sci. Paperbacks.

WECHSLER, D. (1949) Wechsler Intelligence Scale for Children. New York : The Psychological Corporation.

WEINSTEIN, E. and KAHN, R. (1955) Denial of Illness. Springfield, Illinois : Charles C. Thomas.

WEINSTEIN, E., VETTER, R. and SERSEN, E. (1964) Physiological and Experiential Concomitants of the Phantom. New York : Albert Einstein Coll. Med. V.R.A.Proj. No. 427.

WHITEMAN, M. and LUKOFF, I. (1965) Attitudes Toward Blindness and Other Physical Handicaps. J.Soc.Psych., 66, 135-145.

WHITING, J. and CHILD, I. (1953) Child Training and Personality. Yale.

WILLIAMS, P. (1970) Slow Learning Children and Educational Problems. In MITTLER, P. (Ed.) The Psychological Assessment of Mental and Physical Handicaps. London : Methven.

WOLFF, S. (1969) Children Under Stress. Harmondsworth : Penguin Press Ltd.

WRIGHT, B.A. (1960) Physical Disability - A Psychological Approach. New York : Harper and Row.

WRIGHT, E. (1955) The Period of Mourning in Chronic Illness. In HARROWER, M. (Ed.) Medical and Psychological Teamwork in the Care of the Chronically Ill. Springfield, Illinois : Charles C. Thomas.

YARROW, L.J. (1970) Interviewing Children. In MUSSEN, P.H. (Ed.) Handbook of Research Methods in Child Development. New York : Wiley.

YOUNGHUSBAND, E. (1970) Living with Handicap. London : National Bureau for Cooperation in Child Care.

YUGEND, L. (1941) The Role of Parental Attitudes in the Treatment of Diseases in Children. Mental Hygiene, 25, 591-605.

YUKER, H., BLOCK, J. and CAMPBELL, W. (1960) A Scale to Measure Attitudes toward Disabled Persons. Albertson, New York : Human Resources Res.Foundn., Hum.Res. Study No. 5.

YUKER, H., BLOCK, J. and YOUNG, J. (1966) The Measurement of Attitudes toward Disabled Persons. Albertson, New York : Human Resources Center, Hum.Res.Study No. 7.

YULE, W. and RUTTER, M. (1968) Educational Aspects of Childhood Maladjustment : Some Epidemiological Findings Brit.J.Educ.Psych., 38, 1, 7-9.